Undetected Dural Leaks Complicated by Accidental Drainage of Cerebrospinal Fluid (CSF) can Lead to Severe Neurological Deficits

Intrakranielle Hypotension und schwere neurologische Defizite nach akzidenteller Drainage von Liquor bei zuvor undetektierten Duraverletzungen

Abstract

Purpose: Intracranial hypotension has been reported as a complication of accidental drainage after surgical treatment in several cases. Application of negative pressure systems (wound drains, VAC®-therapy, chest tube drainage) had typically led to severe intracranial hypotension including intracranial hemorrhage and tonsillar herniation. In the last year the authors observed 2 cases of accidental spinal drainage of CSF in patients with neurological deficits, regressing after reduction of the device suction.

Material and Methods: We conducted a systematic PubMed-based research of the literature to study the variety and frequency of the reported symptoms from 1st of January 1980 until 1st of October 2015.

Results: Reviewing the literature 24 relevant citations including 27 reported cases of posttraumatic or postoperative loss of CSF leading to neurological symptoms were identified. All 15 reported cases in which a negative pressure suction device had been applied showed severe neurological and radiological symptoms such as coma or brain herniation and intracranial hemorrhage. In all cases patients recovered rapidly after removal of the suction device. Milder symptoms were observed in the patients without negative pressure suction, mainly only presenting with headaches or cranial nerve involvement. Additionally, we give an overview about current recommendations regarding cranial and spinal imaging to rule out dural laceration and cranial hypotension.

Conclusion: Patients with dural laceration complicated by accidental drainage of CSF can present with life-threatening conditions. Increasing use of negative pressure suction devices makes the reported condition an important differential diag-

Zusammenfassung


Schlussfolgerung: Undetektierte Liquorleckagen kompliziert durch akzidentelle postoperative/posttraumatische Drainage von Liquor können zu schweren neurologischen Defiziten bei den betroffenen Patienten führen. Die zunehmende Ver-
wendum von Vakuum-basierten Drainagesystemen führt zu ei-ner hohen Relevanz dieses Krankheitsbildes, welches somit eine wichtige Differentialdiagnose darstellt.

Kernaussagen:
► Undetektierte Liquorleckagen kompliziert durch akzidentelle postoperative/posttraumatische Drainage von Liquor können zu Lebensbedrohlichen Symptomen führen.
► Die zunehmende Verwendung von Vakuum-basierten Drainagesystemen führt zu einer hohen Relevanz der Differentialdiagnose „Liquorunterdrucksyndrom“.

Introduction

Intracranial hypotension may ensue spontaneously after dural puncture or accidental (intraoperative) surgical opening of the dura [1 – 3]. As a complication of this, several cases of accidental drainage after surgical treatment and application of negative pressure systems (wound drain, VAC®-therapy, chest tube drainage) have been reported. Under these circumstances severe intracranial hypotension leading to tonsillar herniation and subdural hemorrhage has been reported [4 – 7]. In the last year the authors noticed 2 cases of accidental drainage of CSF in which patients showed neurological deficits which were regressive after reduction of the device suction. In times of increasing use of negative pressure suction devices, the reported condition presents an important differential diagnosis. To the authors’ knowledge this is the first review that systematically investigates the frequency and variety of emerging symptoms.

Methods


Results

Reviewing the literature, 24 relevant citations including 27 reported cases of posttraumatic or postoperative loss of CSF leading to neurological symptoms were identified (Table 1). All 15 reported cases in which a negative pressure suction device had been applied showed severe neurological symptoms. Milder symptoms were observed in the 10 patients without negative pressure suction. In 2 patients it could not be clearly determined from the literature whether a suction drain had been applied.

Etiology

Fractures of the skull base [8] and the spine [9] can cause traumatic dural tears. In addition spontaneous CSF leaks may occur in approximately 5/100000 patients [2, 3, 10, 11]. If the loss of CSF exceeds its production, intracranial hypotension may ensue [2, 9]. Traumatic dural tears mostly occur after severe spinal trauma. In a retrospective observational study 13% of all patients with traumatic cervical spine injury featured a dural tear [12]. Patients with initial poor neurological state and disruption of the ligamentum flavum were more often affected [11]. Likewise, 25% of lumbar burst fractures exhibited dural tears; all of these patients showed a neurological deficit [12]. Despite the dural tear, none of the studied patients exhibited critical intracranial hypotension [13].

Clinical Presentation

Clinical presentation of intracranial hypotension is heterogeneous [1]. Typical neurological symptoms of CSF overdrainage include nausea, emesis, diplopia, orthostatic headaches, dizziness, difficulties in hearing, visual blurring [2, 10] and cranial nerve palsy [14, 15]. Current publications report on reversible coma, intracranial vasospasms [17] or even stroke and death due to intracranial hypotension [11]. After a loss of CSF of around 10% a dilatation of cerebral sinuses, a sagging of the brain and therefore tension on sensitive vessels, nerves and meninges leads to headaches [18].

Reported complications by application of negative pressure suction devices

Postoperative wound drains

Several reports of patients with severe neurological symptoms after spinal surgery and application of suction drains exist [5, 16, 20 – 24]. An overview of a few cases of postsurgical intracranial hypotension after mainly cranial and only in 2 cases spinal surgery reports about similar symptoms after opening of the dura [19]. Of sixteen patients who remained unconscious or did not become fully responsive after surgery, 4 died and 2 showed brain stem signal intensity changes in MRI. All patients had rapid intraoperative/postoperative CSF loss documented on CT and/or MR imaging studies. The authors came to the conclusion that intracranial hypotension after intracranial and spinal surgery with dural opening is potentially life threatening if patients are left with a subfascial suction drainage.

Similar cases about comatose patients with secondary pseudohy-poxic brain swelling after lumbar laminectomy [5] and after thoracic and lumbar spondylodiscitis surgery have been reported [25]. The initial operative procedure in these cases was extradural. In all cases epidural suction drains (Jackson-Pratt drains) were placed in the tissue bed and the initially oriented patients became comatose. Cranial imaging showed subarachnoid hemorrhage and signs suggestive of cerebral anoxia. The fluid from the drain in one case was positive for β-2 transferrin indicating CSF [5]. This patient fully returned to baseline postoperatively, MRI normalized [5]. The radiographic findings in all cases were re-
Table 1 Patients with postsurgical and posttraumatic intracranial hypotension.

<table>
<thead>
<tr>
<th>postoperative / postoperative, brachial plexus avulsion and reconstructive surgery</th>
<th>suction/ device</th>
<th>symptoms</th>
<th>imaging signs</th>
<th>therapy</th>
<th>outcome</th>
<th>author</th>
</tr>
</thead>
<tbody>
<tr>
<td>pleural drain</td>
<td>postural headaches, headaches worsened after aspiration of the pleural effusion</td>
<td>CT: tonsillar herniation and syringomyelia C1 to C7</td>
<td>surgical closure of the dural tear</td>
<td>headaches immediately resolved, MRI 4 months later: resolution of cerebellar tonsill herniation, regression of the syrinx</td>
<td></td>
<td>Scholsem [26]</td>
</tr>
<tr>
<td>posttraumatic, isolated gunshot wound to the midaxillary line through the tenth intercostal space and a complete spinal cord injury at T12</td>
<td>pleural drain</td>
<td>initially oriented, comatose with complete absence of brainstem reflexes 3 hours after admission</td>
<td>MRI: diffuse cerebral edema, occlusion of the bilateral posterior cerebral arteries, tonsillar herniation</td>
<td>none</td>
<td>death</td>
<td></td>
</tr>
<tr>
<td>postoperative, thoracic laminectomy/fusion</td>
<td>pleural drain</td>
<td>severe neurological deterioration</td>
<td>CT: bilateral subdural hematomas, tonsillar herniation</td>
<td>evacuation of subdural hematomas and bony decompression of the foramen magnum with a cerebellar tonsillectomy</td>
<td>regained full consciousness</td>
<td>Oudeman [23]</td>
</tr>
<tr>
<td>posttraumatic</td>
<td>VAC®</td>
<td>sudden neurological deterioration, coma</td>
<td>narrowing of basal cisterns, tonsillar cerebellar herniation</td>
<td>dural patch</td>
<td>discharge without neurologic sequelae</td>
<td>Sporns [7]</td>
</tr>
<tr>
<td>postoperative, decompressive laminectomies and lumbar sacral arthrodensis L2-S1</td>
<td>subfascial wound drains</td>
<td>severe headache, generalized tonic clonic convolution</td>
<td>CT: acute and chronic subdural and intraventricular hemorrhages</td>
<td>burr hole over the right parietal skull</td>
<td>recovered completely</td>
<td>Huh [36]</td>
</tr>
<tr>
<td>postoperative, posterior lumbar interbody fusion L1-L2</td>
<td>hemovac drain</td>
<td>severe headache with nausea and vomiting</td>
<td>CT: acute subdural hematomas</td>
<td>dural patch</td>
<td>recovered completely</td>
<td>Jung [37]</td>
</tr>
<tr>
<td>postoperative, bilateral L3-L4 interlaminectomies and L3-L4 discectomy</td>
<td>hemovac drain</td>
<td>left side weakness accompanied with nausea and severe headache</td>
<td>CT: acute cranial epidural hematoma</td>
<td>removal of the drain</td>
<td>recovered completely</td>
<td></td>
</tr>
<tr>
<td>postoperative, Laminectomy L4 / L5</td>
<td>Jackson-Pratt drain</td>
<td>abrupt deterioration in mental status</td>
<td>CT: traces of subarachnoid hemorrhage and signs suggestive of cerebral anoxia</td>
<td>removal of the drain</td>
<td>fully recovered on hospital day 10, MRI on hospital day 8 normalized</td>
<td>Fehnel [5]</td>
</tr>
<tr>
<td>postoperative, Harrington rod placement for lumbar scoliosis</td>
<td>Jackson-Pratt drain</td>
<td>abrupt deterioration in mental status</td>
<td>CT: tonsillar herniation with superior cerebellar infarction</td>
<td>removal of the drain</td>
<td>mental status improved slowly after removal of the drain, remained quadriplegic</td>
<td>Andrews [20]</td>
</tr>
<tr>
<td>postoperative, cranio- tommy for resection of cavernous malformation with intraoperative lumbar drainage</td>
<td>lumbar drain</td>
<td>drowsy, opened eyes to commands, oriented only to name</td>
<td>CT: cerebellar tonsillar herniation</td>
<td>lumbar blood patch, decompressive suboccipital craniectomy and C-1 laminectomy with duroplasty</td>
<td>lasting neuropathic pain and cervical cord signal changes on MRI</td>
<td>Sugrue [4]</td>
</tr>
<tr>
<td>postoperative, clipping of a ruptured aneurysm</td>
<td>lumbar drain</td>
<td>coma, signs of cranial nerves involvement, brain stem and cerebellar dysfunction</td>
<td>MRI: brain sagging and cerebellar tonsillar herniation</td>
<td>epidural blood patch, ventricular drainage, and Trendelenburg position</td>
<td>discharged without sequelae</td>
<td>Bloch [21]</td>
</tr>
<tr>
<td>postoperative, lumbar spinal surgery</td>
<td>lumbar drain</td>
<td>neurological deterioration, coma</td>
<td>CT: thrombosis of the superior sagittal sinus, the right transverse sinus, the right sigmoid sinus, and the right jugular vein</td>
<td>dural patch</td>
<td>recovered completely</td>
<td>Lourenço Costa [38]</td>
</tr>
<tr>
<td>tap test with continued CSF flow for almost 30 min</td>
<td>CSF tap</td>
<td>sudden neurological deterioration, pupils sluggishly reacting to light and generalized tonic-clonic seizures</td>
<td>CT: multiple infra- and supratentorial intracerebral hemorrhages with irritation into the ventricular system</td>
<td>ventilatory assistance</td>
<td>death</td>
<td>Ruiz-Sandoval [39]</td>
</tr>
</tbody>
</table>
ported to be most consistent with acute intracranial hypotension relating to acute loss of CSF.

Two patients in the literature presented with neurogenic shock after posterior lumbar interbody fusion (PLIF). One patient showed bradycardia and the other developed cardiac arrest just after the surgical incision was closed and the drainage tube opened. In these cases incarceration of multiple cauda equina rootlets after the accidental dural tear by suction drainage seems to have caused a sudden decrease of cerebrospinal fluid pressure and traction of the cauda equina, which may have led to the vasovagal reflex. This shows that not only cranial but other life-threatening conditions can occur.

<table>
<thead>
<tr>
<th>posttraumatic/postoperative, thoracic spondylodiscitis surgery</th>
<th>epidual suction drain</th>
<th>epileptic seizures, coma</th>
<th>pseudohypoxic brain swelling</th>
<th>removal of the drain</th>
<th>subsequent recovery</th>
<th>Parpaley [22]</th>
</tr>
</thead>
<tbody>
<tr>
<td>postoperative, lumbar spondylodiscitis surgery</td>
<td>epidual suction drain</td>
<td>sudden neurological deterioration, coma</td>
<td>CT: basal ganglia, cerebellar and brainstem infarction</td>
<td>removal of the drain</td>
<td>death</td>
<td>Parpaley [22]</td>
</tr>
<tr>
<td>postoperative, thoracic schwannoma resection</td>
<td>no</td>
<td>intermittent headaches, right ptosis, cranial nerve III palsy</td>
<td>CT: small subdural hematoma MRI: diffuse leptomeningeal enhancement and crowding of the foramen magnum</td>
<td>conservative treatment</td>
<td>fully recovered</td>
<td>Lau [40]</td>
</tr>
<tr>
<td>postoperative, thoracic disc excision</td>
<td>no</td>
<td>acute diplopia, cranial nerve VI palsy</td>
<td>CT: bilateral subdural effusion</td>
<td>conservative treatment</td>
<td>recovered completely in 3 months</td>
<td>Khurana [41]</td>
</tr>
<tr>
<td>postoperative, thoracic disc excision</td>
<td>no</td>
<td>diplopia, occasional headaches, cranial nerve VI palsy</td>
<td>no</td>
<td>conservative treatment</td>
<td>recovered completely over a period of 5 months</td>
<td>Khurana [41]</td>
</tr>
<tr>
<td>postoperative, microscopic discectomy L5-S1</td>
<td>no</td>
<td>delayed postural headache and photophobia</td>
<td>MRI: enhancement of the pachymeninges CT-myelography: CSF leak</td>
<td>epidural blood patch, eventually direct suture plication</td>
<td>discharge without sequelae</td>
<td>Kundu [42]</td>
</tr>
<tr>
<td>postoperative, lumbar surgery for failed back syndrome</td>
<td>no</td>
<td>expressive aphasia, headache</td>
<td>CT: acute subdural hematoma</td>
<td>evacuation of the hematoma, operative dural patch</td>
<td>discharged without sequelae</td>
<td>Sciubba [16]</td>
</tr>
<tr>
<td>postoperative, incidental durotomy</td>
<td>no</td>
<td>–</td>
<td>extensive but reversible cerebral vasospasm</td>
<td>operative dural tear repair</td>
<td>discharged without sequelae</td>
<td>Chaves [17]</td>
</tr>
<tr>
<td>postoperative, spinal instrumentation surgery for scoliosis 3 months prior to her admission</td>
<td>no</td>
<td>headaches</td>
<td>CT: thoracic pedicle screw penetrating and transversing the dura mater at the T3-T4 level</td>
<td>dural patch, revision of the screw</td>
<td>recovered completely</td>
<td>Albayram [43]</td>
</tr>
<tr>
<td>postoperative, surgery for a nerve sheet tumor originating at the level of the right second neural root</td>
<td>no</td>
<td>horizontal diplopia occurred, palsy of the left abducens nerve</td>
<td>cranial MRI: diffuse pachymeningitis thoracic MRI: pseudomeningocele around the second intervertebral foramen</td>
<td>conservative management</td>
<td>complete recovery 6 months later</td>
<td>Bobbio [44]</td>
</tr>
<tr>
<td>postoperative, spinal surgery for severe myelodysplastic scoliosis</td>
<td>no</td>
<td>aphasic, somnolent</td>
<td>CT: massive subdural hematoma, pneumorachis and a malpositioned pedicular screw</td>
<td>cranial decompression and spinal reoperation with dural tear repair</td>
<td>recovered completely</td>
<td>Nowak [45]</td>
</tr>
<tr>
<td>caudal epidural injection</td>
<td>no</td>
<td>orthostatic headache with generalized weakness and syncopal episodes</td>
<td>no</td>
<td>epidural blood patch</td>
<td>recovered completely</td>
<td>Thomas [46]</td>
</tr>
<tr>
<td>postoperative, spondylodiscitis T6/7</td>
<td>–</td>
<td>–</td>
<td>CT: compressed ambient cisterns, basal ganglia hypointensity</td>
<td>–</td>
<td>recovered completely</td>
<td>Hadizadeh [19]</td>
</tr>
<tr>
<td>postoperative, Spondylodiscitis L4/5, spinal fixation</td>
<td>–</td>
<td>–</td>
<td>MRI: brain stem and thalamic/basal ganglia lesions</td>
<td>–</td>
<td>death</td>
<td>Hadizadeh [19]</td>
</tr>
</tbody>
</table>

No = no drain used, – = no information available.

Table 1 (Continuation)
Previously unreported case of a patient who underwent lumbar stabilization. After application of a suction drain the cranial CT shows a frontal hygroma A+B on the right and effacement of the sulci B. After removal of the drain the cranial CT three days later is normal; ventricular size has increased C+D.

Previously reported case of a patient who had severe intracranial hypotension after VAC® wound therapy. Computed tomography (CT) of the head before VAC® application; A Head axial and B Head sagittal MPR showing a normal cranial CT.

Abb. 1 Noch nicht veröffentlichter Fall eines Patienten nach lumbaler, dorsaler Stabilisierung. Nach Anlage einer lumbalen Unterdruckdrainage zeigt das axiale Schädel-CT ein frontales Hygrom rechts A+B und ein Verstreichen der Sulci B. Nach Entfernung der Drainage ist das Kontroll-CT 3 Tage später wieder normal, die Ventrikelweite hat wieder zugenommen C+D.

Abb. 2 Bereits vorab veröffentlichter Fall eines Patienten mit schwerer intrakranieller Hypotension nach Anlage eines VAC-Wundverbandes. Das CT vor VAC-Applikation ist unauffällig A+B.
Report of a new Case
A previously unreported patient underwent lumbar stabilization. A suction drain was applied in the tissue bed and in the postoperative course the patient developed a global aphasia and disorientation. The head CT showed a frontal hygroma on the right and an effacement of the sulci (Fig. 1A, B). The drain was removed immediately and the neurological symptoms of the patient fully recovered on the same day. A control CT three days later was normal (Fig. 1C, D).

VAC® therapy
Recently we have reported about a polytraumatized patient with a life-threatening event of intracranial hypotension after VAC® wound therapy [7]. This patient had suffered a pelvic fracture after a motor vehicle accident. Initial cranial CT was normal (Fig. 2). After application of a VAC® wound dressing, the initially oriented patient became nonresponsive. The following cranial CT showed signs of intracranial hypotension (Fig. 3). Pneumorrhachis in native CT indicated a dural tear (Fig. 4) which was confirmed by CT myelography at L5 / S1 level (Fig. 5) and consequently the VAC® was removed. After application of a dural patch the patient was oriented post-operatively and the cranial CT improved to a normal state (Fig. 6).

Chest tube drainage
Posttraumatic CSF loss with loss of consciousness and tonsillar herniation has been reported as a complication of chest tube drainage after spinal injury [6]. The patient presented to the trauma service awake, alert, and oriented with an isolated gunshot wound leading to a complete spinal cord injury at T12. A chest tube was placed, and the patient was found to be comatose with complete absence of brainstem reflexes 3 hours after admission. MRI and autopsy revealed diffuse cerebral edema, occlusion of the bilateral posterior cerebral arteries, and tonsillar herniation extending several centimeters below the foramen magnum, with petechial hemorrhages and absence of gliosis. Another case of tonsillar herniation and syringomyelia after brachial plexus avulsion and reconstructive surgery with a cerebrospinal fluid leak between the cervical subarachnoid space and the pleural cavity has been reported [26]. The patient showed a persistent right pleural effusion for about four months after reconstructive surgery for a right brachial plexus avulsion. Initially present headaches had worsened considerably after two aspirations of the pleural effusion. MRI demonstrated signs of chronic intracranial hypotension and tonsillar herniation with a presyrinx cavity from vertebral level C1 to C7. Plexus brachial MRI confirmed the presence of a cerebrospinal fluid leak between the avulsed root of C8 and the pulmonary apex. After dural closure, the patient’s headaches immediately resolved, and MRI four months later showed resolution of cerebellar tonsillar herniation and regression of the syrinx.

Radiological Examination

Cranial Imaging
Typical imaging signs of intracranial hypotension which can be depicted in CT and MRI include “slit like” lateral ventricles, subdural fluid collections and effacement of sulci, as well as the downward
transtentorial herniation of the brain with the “sagging brain” appearance [1, 19, 27]. Imaging modality of choice is an MRI of the head. Features include thickening and enhancement of the dura after contrast application, enlargement of venous structures with dilation of the intracranial dural sinuses and spinal epidural plexuses, and engorgement of the pituitary gland [1, 19]. Moreover hypointensities in the thalamus/basal ganglia, brain stem, and cerebellum have been reported as a possible result of impaired drainage through the internal cerebral veins and the vein of Galen into the straight sinus [19, 25].

Subdural hygromas, often bihemispheric and with an infratentorial predominance, usually are named as a typical imaging finding [3, 19]. However, they were only found in 30% of patients and did not correlate with clinical outcome parameters [19].

**Fig. 6** CT-scan head; the sagittal MPR five days after removal of the VAC shows a regressive lowering of the cerebellar tonsils.

**Abb. 6** Das abschließende Schädel-CT fünf Tage nach Entfernung des VAC zeigt eine Zurückverlagerung der Kleinhirntonsillen in die ursprüngliche Position.

**Spinal Imaging**

A possible indication for traumatic dural laceration may be pneumorrhachis, well discernible in CT [13, 28]. With MRI, dural tears can be detected using fluid-sensitive and fat-suppressive sequences. Typical are fluid collections along the paravertebral tissue and spinal nerves [29]. MRI is the modality of choice for depicting plexus injuries and nerve root avulsions which can be associated with dural laceration [30, 31]. For this purpose MR neurography with adjunction of intrathecal contrast medium loss of CSF can directly be detected [7, 34, 35]. However, this contains a risk of herniation or postpunctureal CSF depletion.

**Conclusion**

In most of the cases with severe neurological symptoms, in addition to the dural tear, extra negative pressure had been applied. This implies that apart from the dural tear, additional factors such as suction of cerebrospinal fluid are needed to induce severe symptoms. In conclusion, patients with suspected dural laceration complicated by accidental drainage of CSF can present with life-threatening conditions. Increasing use of negative pressure suction devices makes the reported condition an important differential diagnosis so that acute intracranial hypotension should be considered as an explanation of postoperative coma after cranial or spinal surgery. A precise radiological examination can help to rule out dural laceration and intracranial hypotension.

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Review

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