Persistent cerebrospinal fluid-cutaneous fistula after epidural analgesia: a case report and review of literature

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Abstract

Presented is a case of persistent high-output cerebrospinal fluid (CSF) leak following epidural catheter removal on postoperative day 5 for a 64-year-old patient after exploratory laparotomy. Epidural catheter placement required multiple attempts preoperatively and was complicated by a “wet tap”. Diagnosis of CSF leak was made with glucose, protein and beta-2 transferrin testing. In our review of 30 cases with reported CSF-cutaneous fistula, a trend towards requiring epidural blood patch for symptom relief exists when larger bore needles are used (all 9 reports involving 14 gauge or larger needles and 2 reports of fistulas resulting from a surgical defect) and duration of catheter stay is long. Notably, there were 3 case reports of CSF-cutaneous fistulas occurring without the placement of a catheter. Among 9 cases of fistula reported in the obstetric population, only one patient needed EBP for symptom control. Location of the needle/catheter insertion site at different levels of the neuraxium does not appear to affect the risk of developing a CSF-cutaneous fistula. CSF-cutaneous fistulas are a rare event that should be recognized early due to the potential for infections and neurologic complications. Glucose and protein testing offer a rapid but less specific means of diagnosing a possible CSF leak and can be used as guidance while awaiting results from beta-2 transferrin or beta-trace protein testing for definitive identification of CSF. With literature review, we propose an algorithm to help early detection and management of CSF-cutaneous fistula.

Keywords: Epidural analgesia, cerebrospinal fluid-cutaneous fistula, beta-2 transferrin, postoperative pain management

Introduction

Cerebrospinal fluid (CSF)-cutaneous fistulas are a rare complication of procedures accessing the neuraxium. As such, recognition of this complication may be delayed, and optimal management is unclear. We present a case of an asymptomatic but high-output and prolonged CSF-cutaneous fistula following epidural analgesia for postoperative pain management, as well as a review of the CSF-cutaneous fistula literature. Our patient was managed successfully with conservative measures and an algorithm is proposed to help in early detection and management of CSF-cutaneous fistulas.

Case presentation

A 64-year-old Caucasian female presented with a high-output enterocutaneous fistula and increased herniation of the bowel. The patient had undergone extensive abdominal resection for stage IIIIC serous papillary ovarian adenocarcinoma which was complicated by wound dehiscence treated with vacuum-assisted closure. She subsequently underwent multiple rounds of chemotherapy. On day of surgery, she underwent exploratory laparotomy, lysis of adhesions, takedown of enterocutaneous fistula, small bowel resection, and repair of large incisional hernia using biological mesh (SurgiMend).

Preoperatively, a lumbar epidural catheter was placed for postoperative pain management. Three attempts were made prior to successful epidural placement, each with a 17 gauge Tuohy needle. In the first attempt, the L2-L3 epidural space was found using the loss of resistance technique, but the epidural catheter was unable to be threaded. In the second attempt, the L2-L3 epidural space was again found using the same loss of resistance technique. A Portex epidural catheter was placed, but clear fluid was noted on aspiration and the catheter was removed. The final attempt was successfully completed by a different anesthesiologist at the L1-L2 space and an Arrow epidural catheter was placed with negative aspiration and a negative test dose.

The patient's initial postoperative course was uncomplicated. She was admitted to the surgical intensive care unit (ICU) for close respiratory and hemodynamic monitoring. Upon admission to the ICU, her epidural provided right greater than left sided coverage and required boluses of 5cc of 0.25% bupivacaine followed by 5cc of 0.5% bupivacaine. This resulted in transient leg numbness which resolved spontaneously. Her epidural infusion was started with bupivacaine 0.1% at 12ml/hour in addition to an IV hydromorphone patient-controlled analgesia (PCA) (0.4mg demand dose, 10 minute lockout, no
Although a rare complication of neuraxial procedures, there have been multiple case reports of CSF-cutaneous fistulas. They have occurred following spinal surgery [1-2], long- and short-term intrathecal catheters [3-11], diagnostic and therapeutic lumbar punctures [12], combined spinal and epidurals (CSE) [13], epidural catheters [14-20], and one-shot epidural anaesthetics and steroid injections [21-23]. An overview of CSF-cutaneous fistula reports is presented in Table 1.

In our review of 30 cases with reported CSF-cutaneous fistula, a trend towards requiring EBP for symptom relief exists when larger bore needles are used (all 9 reports involving 14 gauge or larger needles and 2 reports of fistulas resulting from a surgical defect) and duration of catheter stay is long. Notably, there were 3 case reports of CSF-cutaneous fistulas occurring without the placement of a catheter. Among 9 cases of fistula reported in the obstetric population, only one patient needed EBP for symptom control. We do not have a clear explanation for this observation. Location of the needle/catheter insertion site at different levels of the neuraxium does not appear to affect the risk of developing a CSF-cutaneous fistula.

The risk factors hypothesized to contribute to the development of a CSF-cutaneous fistula include steroid use (systemic or local as in the case of epidural steroid injections) [3,21,22], accessing the neuraxium through post-laminectomy scar tissue that may have impaired healing ability [5,21], multiple passes at the same level particularly if using the same needle (even without clear evidence of a “wet tap”) [14,21,23], and duration of subarachnoid drainage and catheter size (in the case of intrathecal catheters) [4,7,12]. It has been suggested that all of these factors might interfere with the healing of a CSF-cutaneous tract. Steroids have a known suppressive effect on wound healing. It has been put forward that multiple passes in the same level with the same needle may lead to fibrin deposition along the tract that may interfere with tract closure [14,23]. If multiple passes are required, it is recommended that the needle be thoroughly cleaned in between passes, a new needle be used, or different levels be attempted [21,23].

In the case of our patient, the multiple attempts at epidural placement, particularly with the history of a “wet tap” may have increased her risk for the development of this fistula.

It is not uncommon for drainage to occur following the removal of an epidural catheter; however, drainage is usually attributed to subcutaneous edema or local anesthetic rather than CSF. It may be that the incidence of CSF leak is under-diagnosed and in fact higher than currently recognized. Given that a delay in management may lead to the development of symptoms of intracranial hypotension or even more serious complications such as subdural hematoma [11] or meningitis [18,22], this is an important entity to be recognized. In neurosurgical/ENT literature, CSF rhinorrhea/otorrhoea is commonly diagnosed by testing for the presence of beta-2 transferrin [13,24] or beta-trace protein [24] in the fluid. Although the beta-2 transferrin assay is highly sensitive and specific for CSF, it can be a time consuming assay. Rapid but less specific tests for CSF that may be useful in these situations.
Cutaneous stitching of the outflow defect has been used to manage CSF pressure and therefore promote fistulous tract healing. Tsai et al. have reported successful and surgical correction have also been reported [21].

The role of antibiotics is unclear. Some reports start empiric antibiotic coverage once a fistulous tract is recognized [20,21]; others forego antibiotics in the afebrile, nontoxic patient [5,23]. One paper suggests the consideration of antibiotics more strongly in the immunocompromised [18], as there have been reports of these fistulous tracts being further complicated by the development of meningitis. Given the

Table 1. CSF-cutaneous fistula literature: summary of reports and diagnostic and treatment approaches.

| Reference | Inciting Event | Needle/Catheter Size | Multiple attempt? | Duration of catheter stay (day) | Diagnostic testing | Antibiotics | PDPH | EBP | Cutaneous stitch |
|-----------|----------------|----------------------|-------------------|-------------------------------|--------------------|-------------|------|-----|-----------------|
| [23]      | Epidural (labor) | INAA                 | Yes               | 0                             | Yes                | None        | Yes  | No  | No              |
| [10]      | IT catheter (labor) | INAA                | No                | INAA                          | None               | None        | Yes  | No  | No              |
| [5]       | IT catheter (surgical anesthesia) | 17G/20G          | No                | <1                            | Yes                | None        | Yes  | No  | Yes             |
| [6]       | IT catheter baclofen pump | 14G/1.6mm      | No                | 8                             | None               | None        | No   | Yes | No              |
| [7]       | IT catheter baclofen pump | 14G               | INAA              | 30                            | None               | None        | Yes  | No  | No              |
| [3]       | Lumbar drain     | 14G                 | INAA              | 5                             | No                 | None        | PDPH | No  | No              |
| [4]       | Lumbar drain     | 14G/16G             | INAA              | 3                             | Yes                | None        | No   | Yes | No              |
| [4]       | Lumbar drain     | 14G/16G             | INAA              | 9                             | Yes                | None        | No   | Yes | Yes             |
| [11]      | Lumbar drain     | INAA                | No                | 3                             | No                 | None        | Yes  | No  | No              |
| [8]       | IT catheter pain pump | INAA            | INAA              | INAA                          | INAA               | INAA        | Yes  | No  | No              |
| [7]       | Lumbar shunt     | INAA                | INAA              | 125                           | No                 | None        | Yes  | No  | No              |
| [9]       | Omaya reservoir  | 2.1mm               | Yes               | 28                            | No                 | None        | No   | Yes | Yes             |
| [21]      | Epidural steroid injection | INAA     | Yes               | 0                             | Yes                | Yes         | Yes  | Yes | Yes             |
| [22]      | Epidural steroid injection | 17G           | Yes               | 0                             | no                 | Yes         | No   | Yes | No              |
| [12]      | Lumbar punctures for chemotherapy | 22G           | yes               | 0                             | no                 | None        | no   | Yes | No              |
| [2]       | laminectomy      | INAA                | INAA              | 0                             | INAA               | INAA        | Yes  | No  | No              |
| [1]       | laminectomy      | INAA                | INAA              | 0                             | INAA               | INAA        | Yes  | No  | No              |

are glucose and protein analysis. Presence of glucose in the leaking fluid would suggest against a leak of epidural medication. Interstitial fluid generally has a one to two order of magnitude greater protein concentration compared to CSF [25,26], therefore a protein concentration ≥1 g/dl would be more suggestive of interstitial fluid leakage and has been used to rule out CSF leakage from epidural sites in the past [25,27]. As the incidence of CSF-cutaneous fistulas is rare, treatment for this complication varies widely and optimal management is unclear. Treatment options have included more conservative measures such as sterile dressings [6,11,19,20,23], fluids, and bed rest in positions to minimize CSF pressure on the presumed dural defect as well as minimize dural traction: lateral decubitus +/- hip flexion, prone +/- a pillow under the abdomen, slight trendelenberg [2,4-6,18,21,23]. Dehydration and glycerol have been trialed [21], presumably to decrease CSF pressure and therefore promote fistulous tract healing. Cutaneous stitching of the outflow defect has been used [4,5,13,21]. Epidural blood patches have been reported to be successful [3,4,6-9,11,12,14,16,20,21] even in cases of fistulas due to surgical defects [1,2]. Subcutaneous infiltration around the fistula with saline has been suggested [4]. And finally, more invasive solutions such as use of fibrin glue, tract scarification and surgical correction have also been reported [2,9,22]. These treatments have been used alone and also in combination.

Our patient was managed successfully with the placement of a cutaneous suture. Presumably, the placement of a suture halts the loss of CSF, and the accumulating CSF in either the epidural or subcutaneous space resulted in a tamponade that stopped further flow from the dural defect, allowing for closure and healing. Despite the large volume of CSF loss (if the approximately 1cc per 3 minutes collected from puncture site was 100% CSF, that would translate to a CSF leak rate of 480cc daily, close to the total daily production of CSF in the average adult), our patient was asymptomatic. Had symptoms occurred, there would have been a lower threshold to hold anticoagulation and attempt epidural blood patching. We would suggest that in the absence of symptoms, a simple sterile cutaneous stitch to prevent external communication with the neuraxium may be all that is required in terms of treatment.

The role of antibiotics is unclear. Some reports start empiric antibiotic coverage once a fistulous tract is recognized [20,21]; others forego antibiotics in the afebrile, nontoxic patient [5,23]. One paper suggests the consideration of antibiotics more strongly in the immunocompromised [18], as there have been reports of these fistulous tracts being further complicated by the development of meningitis. Given the
multiple reports of CSF-cutaneous fistulas being treated in the absence of antibiotics with no infectious complications, we would suggest that antibiotics may not be necessary in the afebrile, nontoxic patient. However, these patients merit a low threshold of suspicion for the initiation of antibiotics should signs and symptoms of localized infection/meningitis appear. A possible algorithm for an approach to suspected CSF-cutaneous fistulas is presented in Figure 1.

Conclusion
CSF-cutaneous fistulas are a rare event that should be recognized early due to the potential for infections and neurologic complications. Glucose and protein testing offer a rapid but less specific means of diagnosing a possible CSF leak and can be used as guidance while awaiting results from beta-2 transferrin or beta-trace protein testing for definitive identification of CSF. Patients who are asymptomatic or contraindicated for invasive procedures may be successfully treated with conservative measures, including cutaneous stitching, alone. Otherwise, epidural blood patching has proved to be an effective technique for the management of these fistulas.

Competing interests
The authors declare that they have no competing interests.

Authors’ contributions

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Figure 1. Algorithm for an approach to suspected CSF-cutaneous fistulas. Recommendations are based on a review of literature and reported diagnostic and treatment approaches.
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