Subcutaneous emphysema and pneumocephalus following Ventriculoperitoneal Shunt (VPS) surgery for hydrocephalus: a case report

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ABSTRACT

Background: Hydrocephalus is still commonly found in pediatric neurosurgery managed by endoscopic third ventriculostomy (ETV) and Ventriculoperitoneal Shunt (VPS). Subcutaneous emphysema and pneumocephalus as a complication of VPS surgery is rare. Subcutaneous emphysema is the presence of air in the skin, while pneumocephalus is the presence of air in the intracranial space. This case study aims to evaluate subcutaneous emphysema and pneumocephalus following ventriculoperitoneal shunt surgery for hydrocephalus.

Case Presentation: We report an 11-month-old girl with hydrocephalus due to aqueductal stenosis who postoperatively had subcutaneous emphysema and pneumocephalus. 1 week after VPS insertion, there was a crepitation under the skin of the neck and chest. The patient occasionally appeared breathless and had a fever. The respiratory rate increased with rales on the right lung, along with the gradually decreased consciousness, and the abdominal wound slowly became dehiscent. There was subcutaneous emphysema along the shunt track and pneumocephalus.

Conclusion: The interest, in this case, could be that of the patient's other morbid conditions, including pneumonia and the dehiscent abdominal wound on the post-VP shunt insertion incision that may have also contributed to causing the subcutaneous emphysema and pneumocephalus.

Keywords: Shunt Complications, Pneumocephalus, Subcutaneous Emphysema, VPS.


INTRODUCTION

Hydrocephalus is defined as an active distension of the brain's ventricular system that results from an inadequate flow of cerebrospinal fluid (CSF) from the point of production in the ventricles into its absorption.1 The causes of hydrocephalus are often divided into congenital or acquired. They may result from various processes such as congenital and malformations, intracranial hemorrhage, infection, trauma, brain tumors, or cysts. Hydrocephalus results from excessive production, flow obstruction, or impaired CSF reabsorption.2 In children, this condition is detrimental due to the expanded ventricles, accompanied by increased CSF pressure, and the flexible skull is enlarged; this compresses and stretches the surrounding brain tissue.3 Despite ETV and shunting being the mainstay treatment for hydrocephalus, its efficacy is still debatable.4 It is well established that shunts have risks of complications that can occur throughout the patient's lifetime, including malfunctions due to obstruction, mechanical disconnection, breakage, infection, or over drainage.2

Subcutaneous emphysema is an accumulation of air in the subcutaneous layer of skin. Skin is structured by the epidermis and dermis, with the subcutaneous tissue beneath the dermis. Injury to the thoracic cavity, sinus cavities, facial bones, barotrauma, bowel perforation, or pulmonary blebs is what usually happens.4,5 Pneumocephalus, the presence of air within the calvarium, as a CSF diversionary procedure complication, is rarely reported. The air may be localized in epidural, subdural, intracisternal, and intraventricular spaces or within potential spaces. The presence of intracranial air after CSF shunting or endoscopic procedures is usually of no particular significance.5 However, tension pneumocephalus represents an infrequent but dangerous complication. Intracranial air results from the reduction of brain volume due to the perioperative CSF leaks during the extrathecal CSF shunt implant or the procedure of ETV.2

Based on those mentioned above, this case study aims to evaluate the hydrocephalus due to aqueductal stenosis who had postoperative subcutaneous emphysema and pneumocephalus. The patient also had pneumonia and abdominal wound dehiscence. This condition is still not commonly reported in the literature.

CASE REPORT

History and Examination
An 11-month-old girl was brought to Dr. Soetemo General Hospital complaining of her head looking larger than other babies her age. There was vomiting, fever, seizures, restlessness, or loss of consciousness. Physical examination reveals a GCS of 15 (E4V5M6), a

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Count of 585, dominant PMN, positive Nonne & Pande, glucose of 43 and protein of 500, which indicates bacterial infection. We then performed EVD. CSF culture showed growth of Pseudomonas aeruginosa (sensitive to amikacin, gentamicin, cefoperazone sulbactam, imipenem, and meropenem).

About 20 days after the insertion of EVD and antibiotics administration, routine CSF analysis showed normal results. Gram staining and culture did not show bacterial growth. The patient’s condition was also stable without any complaints, with a GCS of 15. We performed VP Shunting on the right side. There were no complications during the surgery.

About 1 week after shunt insertion, there was a crepitation under the skin of the neck and chest. The patient occasionally appeared breathless and had a fever. The respiratory rate increased with rales on the right lung and gradually decreased consciousness. The abdominal wound slowly became dehiscent (Figure 3). We found E. coli from the wound bed culture. We performed head CT (Figure 4) and Chest imaging (Figure 5). Chest X-rays and CT scan revealed pneumonia and subcutaneous emphysema in the right chest and neck. A head CT scan showed intraventricular pneumocephalus in both lateral ventricles. Shunt exteriorization was performed. 2 days later, due to persisting pneumocephalus and a CSF analysis indicating an infection, we removed the shunt and did EVD insertion and debridement of the abdominal wound.

After several days of treatment, the patient’s pneumonia got worse. The breathlessness worsened and consciousness was decreased, along with vital signs deterioration. The patient fell into a respiratory failure, aggravated by the patient’s septic state. The patient became restless, suffered cardiac arrest and unfortunately died.

DISCUSSION

The incidence of subcutaneous emphysema and pneumocephalus post-VP shunt insertion is rare. This could manifest immediately after the procedure or in a delayed fashion. Immediate postoperative pneumocephalus following shunt procedures occurs due to excessive

Figure 1. Head T1-weighted MR images in sagittal (A) and axial (B) planes obtained at initial presentation revealed triventricular ventriculomegaly with a normal-sized 4th ventricle consistent with aqueductal stenosis.

Figure 2. Axial Head CT non-contrast (A) and contrast (B) revealed lateral ventriculomegaly with leptomeningeal enhancement and showed contrast enhancement layering occipital horn (red arrow) at left and right ventricles.

Figure 3. Dehiscence wound (red arrow) from previous VP shunt insertion.

symmetrical pupil with positive light reflex in both eyes and visibly frontal bossing and sunset phenomenon with an FOC of 50cm (>2SD). Laboratory results were within normal limits. MRI revealed both lateral and 3rd dilation, suggesting non-Communicating hydrocephalus with aqueductal stenosis ventricles (Figure 1). ETV’s success score was 70. The patient had received ETV and CPC; there were no complications during the surgery.

About 2 days later, the patient had a fever (38C), decreased consciousness with a GCS of 12 (E3V4M5), and vomiting. CT scan with contrast revealed hydrocephalus with an impression of meningoencephalitis and ventriculitis (Figure 2). CSF study revealed a yellowish color, increased cell
drainage of the CSF during insertion of the ventricular end of the shunt leading to decreased (ICP). The non-compliant ventricular system does not collapse after excessive drainage of CSF, leading to the influx of air into the ventricular system. On the other hand, delayed pneumocephalus develops due to air leakage from a defect in the skull base or a scalp ventricle fistula not found in this patient. Air can be trapped immediately post-VPS surgery during tunneling for abdominal drain insertion and accumulate under the skin along the shunt tract. We believe other things cause this accumulation of air because it doesn’t appear directly after VPS surgery.

In this case, the particular interest could be that of the patient’s other morbid conditions, including pneumonia and the dehiscent abdominal wound on the past VP shunt insertion incision. We suspect these conditions could be the risk factors for developing subcutaneous emphysema and pneumocephalus. The mechanism of early or immediate pneumocephalus associated with CSF leaks is known as the “inverted bottle.” When CSF flows out, a negative pressure is created within the skull and the space made gets filled with air. Most experts agree that the initial step in a tension pneumocephalus is removing intracranial air. In cases with a defect at the skull base, the treatment of choice is sealing the defect through an open or endoscopic method. However, when the origin of the pneumocephalus is unknown, like in our case, the approach was first to remove the intracranial air; we did EVD insertion.

There was also spontaneous subcutaneous emphysema in the neck region and right hemithorax, and the patient could not pinpoint how this occurred. In this case, we suspect that the subcutaneous emphysema could contribute to the development of the pneumocephalus due to its presence in the proximity of the shunt tract. Air could extend into the scalp and slowly infiltrate the intracranial from the defect present after the VP shunt insertion. We suspect the subcutaneous emphysema is related to pneumonia or an infected abdominal wound.

Several cases have reported subcutaneous emphysema as a complication of pneumonia. A previous study reported the occurrence of subcutaneous emphysema in Staphylococcal pneumonia, measles, Pneumocystis carinii infection, influenza pneumonia and pertussis, especially among children. The previous study has considered the cause of non-traumatic subcutaneous emphysema to be caused by weakness of either the alveolar or bronchial wall. Increased intrapleural pressure following excessive and prolonged coughing leads to rupture at a weakened point allowing air to escape from the tissue. Air escapes via peribronchial or perivascular channels to the mediastinum, spreads into loose alveolar tissue, and gains entry to the neck. Subcutaneous emphysema in the absence of pneumomediastinum or pneumothorax is unusual. In this case, we have looked into the possibility of pneumomediastinum or pneumothorax, but none were visible on the Thorax CT Scan performed on this case. We feel that this case is unusual and could be of particular interest to be looked into.

We also believe the possibility that the emphysema is due to gas-forming organisms, based on previous reports of gas gangrene (Clostridial myonecrosis) or crepitant infection caused by other gas-forming bacteria, such as anaerobic streptococci and some coliform bacteria. We obtained E. coli growth on the wound bed culture. A decision for surgical debridement was made. From

**Figure 4.** Noncontrast Head CT images, sagittal (A) and axial (B) showing lateral ventriculomegaly with pneumoventricle (red Arrow). (10 days after shunt implantation).

**Figure 5.** Chest X-ray (A) and CT Scan Thorax (B) after shunt implantation. (A) Pneumonia with lucent area in the neck soft tissue to the right and left hemithorax, indicating subcutaneous emphysema. (B) Subcutaneous emphysema (red arrow) on the right hemithorax region.
debridement, findings showed no pus and no intraperitoneal fistule. In this case, we hypothesized that abdominal wounds that become dehiscent can be caused by subcutaneous emphysema that forms before and extends to the peritoneal shunt tract, exerts pressure and causes ischemic injury to the abdomen, or vice versa. Infection in the abdomen wound causes gas to form, so extending to the thorax, colli, and intracranial space (pneumocephalus).11-15

Limitations that apply to our study include a limited study regarding subcutaneous emphysema and pneumocephalus post-VP shunt insertion. The mechanism of the complication has not yet been fully understood. We hope further research will shed light on this phenomenon in the future.

CONCLUSION
In the case of subcutaneous emphysema and pneumocephalus post-VP shunt surgery, several factors can cause it to occur. No theory or literature explains this in detail, but we must still evaluate the presence of other pathological factors that might cause these conditions. Factors that may be contributing to iatrogenic pneumocephalus, such as site and duration of intracranial surgery; the presence of gross hydrocephalus, a functioning shunt or CSF drain, or a CSF fistula; intraoperative administration of mannitol; and nitrous oxide anesthesia should also be considered. Surgical decision-making in pneumocephalus depends on the presence of tension or no tension pneumocephalus. In this case, we also decided to do shunt removal and switch to EVD due to tension pneumocephalus and ventriculitis.

CONFLICT OF INTEREST
The authors declare that there is no conflict of interest regarding the manuscript.

ETHICAL CONSIDERATION
Not applicable as no patient identity was disclosed. However, this case study follows ICMJE and COPE protocols for publication ethics.

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AUTHOR CONTRIBUTIONS
Muhammad Wildan Hakim contributed to the study’s conceptualization, data collection, writing, and editing. Muhammad Arifin Parenrengi and Wihasto Suryaningtyas contributed to reviewing, editing, and finalizing the manuscript of the study.

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