

[Case report]

## Recurrent Shoulder Tip Pain After Ventriculoperitoneal Shunt Placement Associated with Infectious Peritonitis with *Propionibacterium acnes*; A Case Report and Review of the Literature

Kohei SUZUKI<sup>1\*</sup>, Takeshi SAITO<sup>1</sup>, Kyohei SAKAI<sup>1</sup>, Tadashi MIYAGAWA<sup>2</sup>, Yuko HONDA<sup>3</sup>, Takayuki HOSHINA<sup>3</sup>, Masato OGAWA<sup>3</sup>, Hiroshi ASAI<sup>3</sup>, Takeshi YAMANOCHI<sup>4</sup> and Junkoh YAMAMOTO<sup>1</sup>

<sup>1</sup> Department of Neurosurgery, School of Medicine, University of Occupational and Environmental Health, Japan. Yahatanishi-ku, Kitakyushu 807-8555, Japan

<sup>2</sup> Department of Pediatric Neurosurgery, Children's Medical Center, Matsudo General Medical Center, Japan. Sendabori, Matsudo 207-2296, Japan

<sup>3</sup> Department of Pediatrics, School of Medicine, University of Occupational and Environmental Health, Japan. Yahatanishi-ku, Kitakyushu 807-8555, Japan

<sup>4</sup> Department of Pediatric Surgery, School of Medicine, University of Occupational and Environmental Health, Japan. Yahatanishi-ku, Kitakyushu 807-8555, Japan

**Abstract :** Ventriculoperitoneal (VP) shunt placement is commonly performed for the treatment of hydrocephalus, and several complications of this procedure are well known. Radiating shoulder tip pain after VP shunt placement has been reported as an unusual complication in a few cases, associated with dislocation of the peritoneal catheter. We described the case of a 9-year-old girl who presented with recurrent radiating shoulder tip pain after VP shunt placement. The pain recurred after peritoneal catheter repositioning because of peritoneal inflammation and adhesion due to peritonitis with *Propionibacterium acnes* (*P. acnes*). This bacterium was isolated using 16S ribosomal RNA gene polymerase chain reaction (16S rRNA gene PCR), and anaerobic and prolonged culture tests. After antibacterial treatment, ventriculoarterial (VA) shunt placement was successfully performed. Hemidiaphragm irritation by the peritoneal catheter leads to radiating shoulder tip pain, and peritoneal inflammation and adhesion caused by infectious peritonitis may cause recurrence of this despite catheter repositioning. Clinicians should be aware of shoulder pain as a complication of VP shunt placement, and should consider VA shunt placement as an alternative treatment if this symptom recurs after catheter repositioning. Furthermore, 16S rRNA gene PCR and anaerobic and prolonged culture tests should be considered to detect *P. acnes* infection.

**Keywords :** shoulder pain, ventriculoperitoneal shunt, ventriculoarterial shunt, *propionibacterium acnes*, pediatric neurosurgery.

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\*Corresponding Author: Kohei SUZUKI, Department of Neurosurgery, School of Medicine, University of Occupational and Environmental Health, Japan. Yahatanishi-ku, Kitakyushu 807-8555, Japan, Tel: +81-93-691-7257, Fax: +81-93-691-8799, E-mail: s-kohei@clnc.uoeh-u.ac.jp

## Introduction

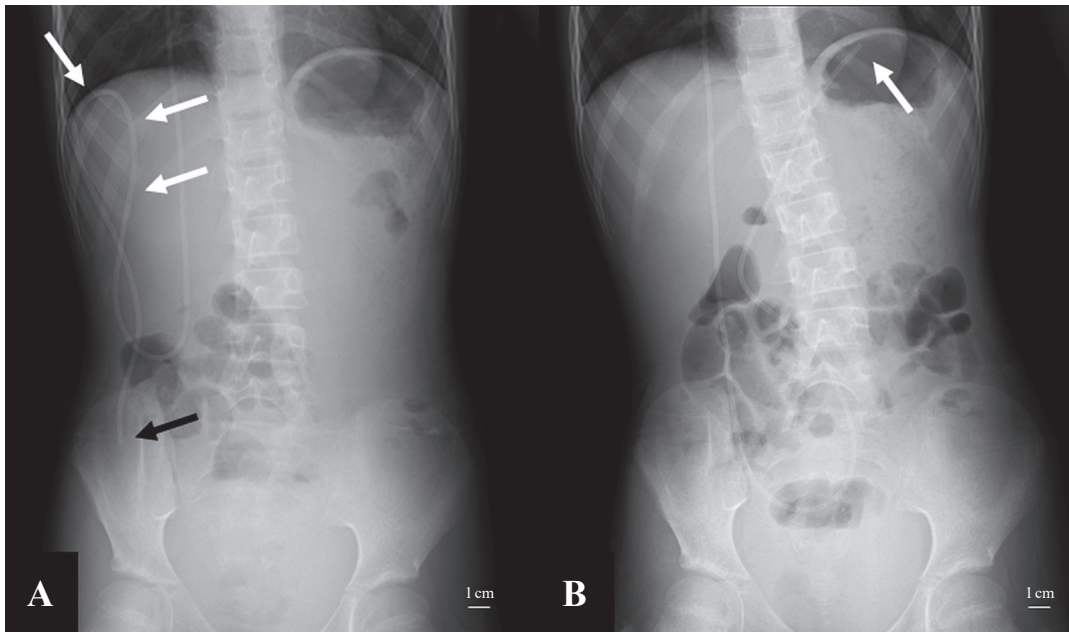
Cerebrospinal fluid (CSF) shunt placement, most commonly involving a ventriculoperitoneal (VP) shunt, is the standard procedure used for the treatment of chronic hydrocephalus. Morbidity and mortality of shunt placement have been reported to range from 23.5 to 29.0% and 3.0 to 5.8%, respectively. Furthermore, the rate of shunt revision has been reported to range from 21.6 to 35.1% [1–3]. Complications such as shunt infection and obstruction, mechanical shunt failure, over drainage, and abdominal complications are encountered with higher frequency. However, a few cases of unusual complications associated with dislocation of the peritoneal catheter, resolved by repositioning of the shunt catheter, have been reported [4–6].

Here, we describe a 9-year-old girl who presented with recurrent shoulder tip pain as a VP shunt complication. This was attributed to bacterial peritonitis with *Propionibacterium acnes* (*P. acnes*) and was resolved after successful ventriculoarterial (VA) shunt placement.

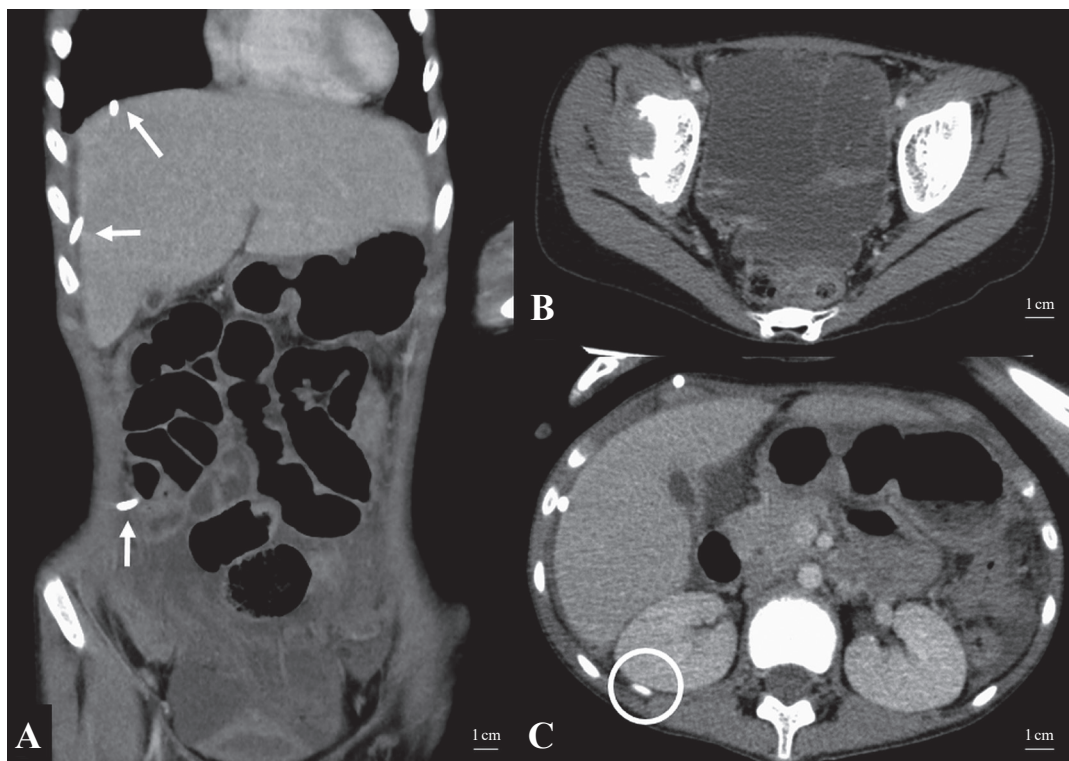
## Case Report

A 9-year-old Japanese girl presented with intermittent abdominal pain which started six days prior and was gradually worsening. She was admitted in July 2018 to the University of Occupational and Environmental Health Hospital in Kitakyushu, Japan. She had a background history of obstructive hydrocephalus caused by a hypothalamic pilocytic astrocytoma, and was treated with VP shunt placement five months prior. After her admission, a clinical examination showed right lower abdominal and hypochondriac pain and radiating pain to her right shoulder. Redness and pain along the shunt catheter were not observed. Serological examination revealed an elevated C-reactive protein (CRP) level (9.96 mg/dl) and a peripheral white blood cell count of 7,900 cells/mm, with 77.7% neutrophils. A CSF culture test was negative. An abdominal X-ray showed that the peritoneal catheter was wrapped over the front of the liver and the catheter tip was located in the right lower abdomen, and these findings were consistent with the site of pain (Fig. 1A). Enhanced abdominal computed tomography (CT) revealed a cystic

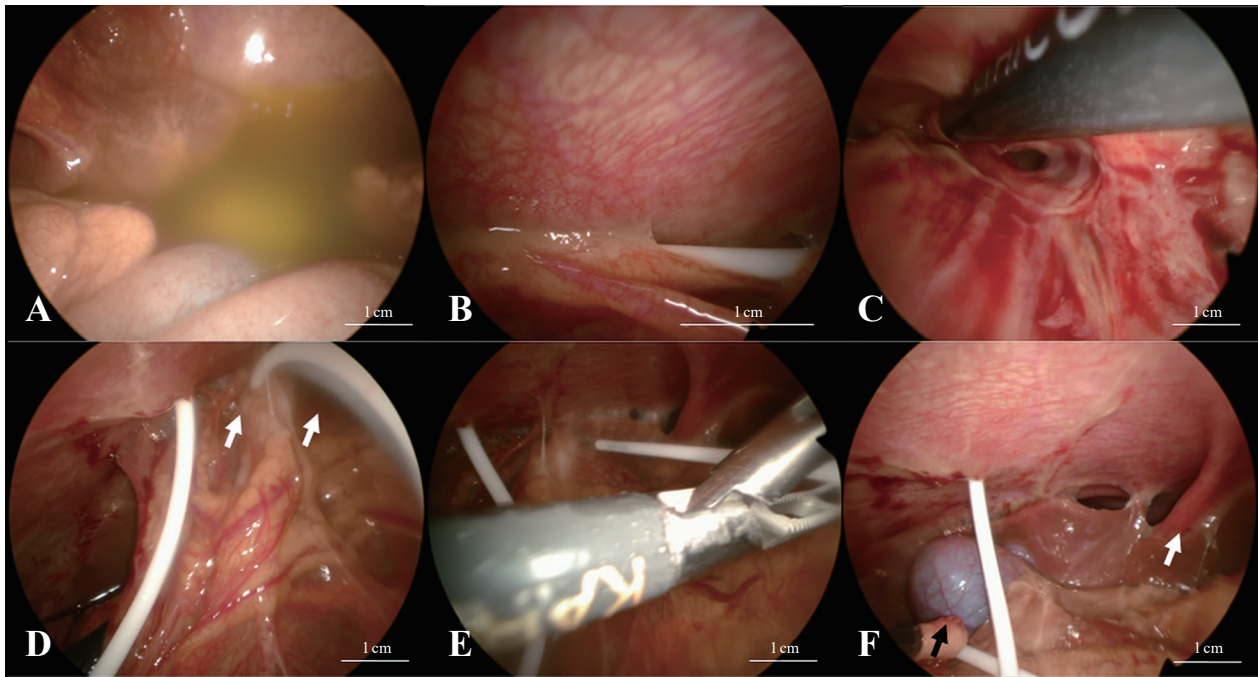
lesion in the pelvis accompanied by intestinal edema. A lumen-like structure was found to be connected to the catheter tip (Fig. 2A–C). Abdominal pain without fever elevation persisted, and the pain changed position daily. Follow-up serological examination showed that CRP level remained elevated (13.11 mg/dl). Antibiotic therapy with tazobactam/piperacillin (TAZ/PIPC) was initiated based on suspected infectious peritonitis. Despite antibiotic therapy, the patient's abdominal pain and serological status did not improve. She presented with left hypochondriac and radiating shoulder tip pain five days after her admission. A follow-up abdominal X-ray showed the peritoneal catheter tip was located just under the left hemidiaphragm, and this was consistent with left hypochondriac pain (Fig. 1B). Because of the observed clinical course, the abdominal and radiating shoulder pain were thought to be associated with the peritoneal catheter. Seven days after her admission, peritoneal catheter repositioning assisted by laparoscopy was performed. Laparoscopy findings showed extensive adherence of the peritoneum and greater omentum, and cloudy ascitic fluid in the pouch of Douglas. Particularly around the catheter, there was peritoneal redness and rigid adherence (Fig. 3A–F). An intraoperative smear test was negative for bacteria, so aseptic peritonitis was diagnosed. Repositioning of the peritoneal catheter and replacement with a silicone catheter (Medtronic, CA, USA) were performed, given the possibility of an allergic reaction to the catheter. Aerobic bacterial culture tests of the CSF and ascitic fluid were negative. After the operation, a patch test was performed, in which an allergic reaction to the shunt catheter was not confirmed. Her clinical symptoms and serological status improved immediately following the operation, and she was discharged eight days later. However, *P. acnes* was isolated from the CSF and ascitic fluid ten days after the operation by anaerobic and prolonged culture tests. Moreover, 16S ribosomal RNA gene polymerase chain reaction (16S rRNA gene PCR) of ascitic fluid obtained in the operation also isolated *P. acnes* [7]. Because she had not presented with a fever, we first considered that this was a result of contamination during the operation. However, abdominal pain recurred four days after her discharge, and this symptom gradually worsened. She was admitted to our hospital seven days



**Fig. 1. A: Initial abdominal X-ray findings.** An abdominal X-ray showed that the peritoneal catheter was wrapping over the front of the liver and attached just under the right hemidiaphragm (white arrows) and the catheter tip was located in the right lower abdomen (black arrow). **B: Followed-up abdominal X-ray findings.** A follow-up abdominal X-ray showed that the peritoneal catheter was curving upwards and the catheter tip was located just under the left hemidiaphragm (white arrow).



**Fig. 2. A: Location of the peritoneal catheter.** An enhanced abdominal CT showed that the peritoneal catheter was attached to the right hemidiaphragm beside the liver and the catheter tip was located in the right lower abdomen. **B: Abdominal cystic lesion.** A cystic lesion in the pelvis was accompanied by intestinal edema. **C: Lumen-like structure.** A lumen-like structure was connected to the catheter tip (white circle).



**Fig. 3. Laparoscopy findings.** **A: Ascitic fluid.** Slightly cloudy ascitic fluid in the pouch of Douglas. **B: Adherence and redness of peritoneum.** Adherence and redness of the peritoneum and greater omentum, with rigid adherence around the catheter. **C: Detachment of the adherence.** Detachment of the adherence by forceps. **D, E: Pull out the catheter.** Distal peritoneal catheter wrapping over to the liver (arrow) and pulled out from rigid adherence gently by forceps. **F: After detachment of the adherence.** The bottom side of the liver, round ligament of the liver (white arrow), and gallbladder (black arrow) were observed following detachment of the adherence around the catheter tract.

after her initial discharge. On admission, colicky abdominal pain was observed, followed by headache and mild consciousness disturbance. Head and abdominal CT scans showed hydrocephalus, and obstruction of the abdominal catheter by adhesion tissue was suspected. Symptoms improved transiently after adjustment of the shunt pressure setting, but then recurred and worsened. Abdominal and radiating shoulder tip pain consistent with the peritoneal catheter also recurred. Anaerobic and prolonged culture tests once again isolated *P. acnes* in CSF from the shunt valve. An external drainage operation was performed eleven days after her second admission. The antibiotics were changed to ceftriaxone sodium (CTRX) according to the results of bacterial susceptibility testing. After adequate antibiotic therapy, her general and serological status improved. Subsequently, VA shunt placement was performed, and her abdominal and shoulder tip pain were immediately resolved. No recurrence has been noted in the year following this procedure.

## Discussion

Several complications following VP shunt placement are well known and can occur anywhere along its course from the cerebral ventricles to the peritoneal cavity. Shunt infection is the most commonly observed complication, with a reported frequency of 8.8 to 12.8% in pediatric patients [8]. Abdominal complications also occur in 21.0 to 22.9% of patients [9, 10]. Clinical presentation is dependent on the site of infection. The occurrence of fever at some point in a patient's hospitalization is the most frequently recorded symptom, and this has been observed in 78% of cases of shunt infection [11]. Vomiting, a CSF leak, purulence, skin erosion, meningismus, erythema, and abdominal pain are all recognized signs of a CSF shunt infection. Radiating shoulder tip pain is a rare complication associated with VP shunt placement, which, to the best of our knowledge, has only been reported in five cases (Table 1) [4-6]. Those pain has been shown to occur at a relatively young age, and on the same side

**Table 1. Summary of shoulder tip pain after VP shunt placement**

Reference	Case	Age	Sex	duration from diagnosis	Side of the shoulder pain	location of the tip of the peritoneal catheter	Treatment	Infection	Adhesion	Prognosis
Tubbs RS, <i>et al.</i> 2005 [6]	1	10	F	4 months	R	R	shunt revision	NL	NL	resolved
	2	12	F	4 months	R	R	shunt revision	NL	NL	resolved
	3	16	M	4 months	R / R	R / R	shunt revision	NL	NL	recurrence / resolved
Lim C, <i>et al.</i> 2005 [5]	4	20	M	3 weeks	L	L	catheter repositioning	NL	-	resolved
Lang JL, <i>et al.</i> 2011 [4]	5	29	F	1 year	R	R	catheter repositioning	-	NL	resolved
Present case	6	9	F	1 week	R / L	R / L	VA shunt	+	+	recurrence / resolved

VP: ventriculoperitoneal, F: female, M: male, R: right, L: left, NL: not listed, VA: ventriculoarterial

that the peritoneal catheter tip is located. In most observed cases, diagnosis took over a month. Shoulder tip pain may be under-recognized as a complication of VP shunt placement because it is rare and can be caused by several other medical conditions, such as cardiac and gallbladder problems.

The pathogenesis of shoulder tip pain after VP shunt placement is thought to involve diaphragmatic irritation by the migrated peritoneal catheter, and this is seen via laparoscopy in 35–63% of cases [12, 13]. The diaphragm, which acts to separate the thoracic and abdominal cavities, derives from the fourth cervical mesoderm and its innervation descends from the cervical to the lower thoracic regions [5]. In the present case, peritoneal catheter movement was disturbed, and the tip of the peritoneal catheter was fixed in the left side with rigid abdominal adherence caused by infectious peritonitis with *P. acnes*. In addition, laparoscopic findings confirmed that the peritoneal catheter was wrapping over just under the diaphragm with rigid adherence.

Treatment of shoulder pain after VP shunt placement is performed via shunt revision or peritoneal catheter repositioning. Previous reports described that this immediately resolved pain in all cases [4–6]. In one case, shoulder tip pain resolved spontaneously after a few weeks of observation, but this recurred and required subsequent peritoneal catheter repositioning [6]. In the present case, shoulder tip pain was resolved im-

mediately after peritoneal catheter repositioning, but the pain recurred a few days following this operation. Laparoscopic and pathological findings showed intra-peritoneal inflammatory changes including adhesion of the peritoneum and greater omentum, and bacterial peritonitis with *P. acnes* was diagnosed by 16S rRNA gene PCR and a prolonged culture test. From these findings, we considered that abdominal adherence disturbing peritoneal catheter movement may have led to the observed repeated shoulder tip pain. Insertion of a new peritoneal catheter could have led to further inflammatory changes and pain recurrence due to irritation of the diaphragm. Therefore, externalization of the peritoneal catheter was first performed, followed by VA shunt placement after adequate medication. Given that inflammation and adherence of the abdominal cavity may affect the recurrence of shoulder tip pain after VP shunt placement, clinicians should consider not only catheter repositioning but also VA shunt placement as alternative treatments.

Several studies have described the pathogens involved in CSF shunt infection and have identified coagulase-negative staphylococcus (CoNS) and *Staphylococcus aureus* as the two predominant etiologic agents. CoNS has been shown to be the most common, accounting for nearly 75% of CSF shunt infections, followed by gram-negative bacteria [8]. Recently, the increased use of anaerobic cultures and prolonged incubation times has led to an increased recognition of

*P. acnes* as an additional infection-causing pathogen. *P. acnes* is an anaerobic, Gram-positive, bacillus bacterium which has been isolated in approximately 15% of cases of CSF shunt infection [14]. In pediatric patients, *P. acnes* is most commonly isolated in distal catheter infections occurring in those aged between 7 and 15. Shunt infections with *P. acnes* usually present with a mild clinical course, and in some cases have been shown to cause pseudocyst formation [8, 14]. Some reviews have shown that *P. acnes* infection did not result in the development of fever, while other studies did not routinely test for this pathogen [11]. *P. acnes* is notoriously difficult to culture using standard aerobic techniques, so anaerobic culture tests and prolonged incubation times have greatly improved the detection of this bacteria in shunt infections [7, 15]. PCR is also described as an effective method to detect CSF shunt infection with *P. acnes*. Broad-range 16S rRNA gene PCR identifies the 16S protein of rRNA that is present in almost all bacteria. The rRNA sequences obtained are then compared with sequences of the databases and the bacterial species identified [16]. This analysis technique identifies *P. acnes* rapidly with high sensitivity [7]. In the present case, infectious peritonitis with *P. acnes* was considered to be the cause of the recurring shoulder tip pain. The distinction between infection and contamination remains difficult in cases with no fever [14], however clinicians should be aware of the possibility of *P. acnes* CSF shunt infection, especially in pediatric patients. Moreover, additional tests, such as 16S rRNA gene PCR and anaerobic culture tests, should be considered in cases of suspected CSF infection, with or without fever.

### Conclusion

Shoulder tip pain following VP shunt placement may be under-recognized as a complication, and clinicians should be aware of this. In most cases, peritoneal catheter repositioning is effective, however abdominal inflammation and adhesion may result in recurrence of this symptom. Therefore, the rapid detection of shunt infection is important for the resolution of clinical symptoms. 16S rRNA gene PCR and anaerobic culture tests should be considered even if the clinical course appears mild and no fever elevation

is observed. In cases of shoulder tip pain recurrence following peritoneal catheter repositioning, VA shunt placement should be considered as an alternative treatment when the infection is completely resolved.

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### Conflict of Interest

The authors declare that they have no conflict of interest.

### References

1. Hanlo PW, Cinalli G, Vandertop WP *et al* (2003): Treatment of hydrocephalus determined by the European Orbis Sigma Valve II survey: A multicenter prospective 5-year shunt survival study in children and adults in whom a flow-regulating shunt was used. *J Neurosurg* 99: 52–57
2. Pollack IF, Albright AL & Adelson PD (1999): A randomized, controlled study of a programmable shunt valve versus a conventional valve for patients with hydrocephalus. Hakim-Medos Investigator Group. *Neurosurgery* 45: 1399–1408; discussion 1408–1411
3. Wu Y, Green NL, Wrensch MR, Zhao S & Gupta N (2007): Ventriculoperitoneal shunt complications in California: 1990 to 2000. *Neurosurgery* 61: 557–562; discussion 562–563
4. Lang JL, Emelifeonwu JA, Hatfield RH & Leach P (2011): Misdiagnosed shoulder tip pain: Complication of a ventriculoperitoneal shunt. *Acta Neurochir (Wien)* 153: 1715–1716
5. Lim C & O'Sullivan MG (2005): Shoulder tip pain: An under-reported complication of ventriculoperitoneal shunt. *Br J Neurosurg* 19: 354–356
6. Tubbs RS, Wellons JC 3rd, Blount JP, Grabb PA & Oakes WJ (2005): Referred shoulder pain from ventriculoperitoneal shunts. Report of three cases. *J Neurosurg* 102 (2 Suppl): 218–220
7. Banks JT, Bharara S, Tubbs RS, Wolff CL, Gillespie GY, Markert JM & Blount JP (2005): Polymerase chain reaction for the rapid detection of cerebrospinal fluid

- shunt or ventriculostomy infections. *Neurosurgery* 57: 1237–1243; discussion 1237–1243
8. Adams DJ & Rajnik M (2014): Microbiology and treatment of cerebrospinal fluid shunt infections in children. *Curr Infect Dis Rep* 16: 427
  9. Chung JJ, Yu JS, Kim JH, Nam SJ & Kim MJ (2009): Intraabdominal complications secondary to ventriculoperitoneal shunts: CT findings and review of the literature. *AJR Am J Roentgenol* 193: 1311–1317
  10. Pandey A, Gangopadhyay AN, Sharma SP, Upadhyaya VD, Kumar V, Gopal SC, Gupta DK & Srivastava A (2009): Placement of the peritoneal end of a ventriculoperitoneal shunt in the suprahepatic space: Does it improve prognosis? *Pediatr Neurosurg* 45: 6–10
  11. Zervos T & Walters BC (2019): Diagnosis of ventricular shunt infection in children: A systematic review. *World Neurosurg* 129: 34–44
  12. Dobbs FF, Kumar V, Alexander JI & Hull MG (1987): Pain after laparoscopy related to posture and ring versus clip sterilization. *Br J Obstet Gynaecol* 94: 262–266
  13. Sarli L, Costi R, Sansebastiano G, Trivelli M & Roncoroni L (2000): Prospective randomized trial of low-pressure pneumoperitoneum for reduction of shoulder-tip pain following laparoscopy. *Br J Surg* 87: 1161–1165
  14. Aubin GG, Portillo ME, Trampuz A & Corvec S (2014): *Propionibacterium acnes*, an emerging pathogen: From acne to implant-infections, from phylotype to resistance. *Med Mal Infect* 44: 241–250
  15. Arnell K, Cesarini K, Lagerqvist-Widh A, Wester T & Sjölin J (2008): Cerebrospinal fluid shunt infections in children over a 13-year period: Anaerobic cultures and comparison of clinical signs of infection with *Propionibacterium acnes* and with other bacteria. *J Neurosurg Pediatr* 1: 366–372
  16. Carneiro IM, Pereira AS, Pinto S, Prata F, Faria CC & Marques JG (2018): *Propionibacterium acnes*: Cause of cerebrospinal fluid shunt infection. *Pediatr Infect Dis J* 37: e168–e169
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*Propionibacterium acnes*関連腹膜炎によりVPシャント後に繰り返し肩への放散痛を呈した一例

鈴木 恒平<sup>1</sup>, 齋藤 健<sup>1</sup>, 酒井 恭平<sup>1</sup>, 宮川 正<sup>2</sup>, 本田 裕子<sup>3</sup>, 保科 隆之<sup>3</sup>, 小川 将人<sup>3</sup>,  
浅井 完<sup>3</sup>, 山内 健<sup>4</sup>, 山本 淳考<sup>1</sup>

<sup>1</sup>産業医科大学 医学部 脳神経外科学

<sup>2</sup>松戸市立総合医療センター 小児脳神経外科

<sup>3</sup>産業医科大学 医学部 小児科

<sup>4</sup>産業医科大学 医学部 小児外科

**要 旨**：脳室腹腔(VP)シャント術は水頭症に対して一般的に行われ、手術に伴ういくつかの合併症についてもよく知られている。一方、VPシャント後に迷入した腹側カテーテルによる横隔膜の刺激に伴う肩への放散痛は稀な合併症として数例の報告があるのみである。今回、*Propionibacterium acnes*(*P. acnes*)関連腹膜炎による腹膜炎と癒着のため、VPシャント再建術後にも繰り返し肩への放散痛を呈した9歳女児の症例を経験した。*P. acnes*は一般培養検査で検出されず、16SリボソームRNA遺伝子ポリメラーゼ連鎖反応(16SrRNA遺伝子PCR)および嫌気性培養、増菌培養で検出され、十分な抗生剤治療の後、脳室心房(VA)シャント術を施行し、その後肩への放散痛は再燃なく経過している。腹側カテーテルによる横隔膜への器械的な刺激は肩への放散痛を引き起こし、カテーテルの入れ替えによって改善するとされるが、感染性腹膜炎による腹膜炎や癒着はカテーテルの入れ替え後も症状を再燃させる因子になる可能性がある。稀ながら腹側カテーテルによる肩への放散痛はVPシャントの合併症として認識する必要がある。また、カテーテル交換後も症状再燃を繰り返す際には、弱毒菌による感染症の除外のためPCRや嫌気・増菌培養を行い、VAシャントへの切り替えを検討する必要がある。

**キーワード**：肩放散痛, VPシャント, VAシャント, *Propionibacterium acnes*, 小児脳神経外科.