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Silicone hypersensitivity necessitating removal of ventriculoperitoneal shunt in schizencephaly patient complicated with hydrocephalus: A case report

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ABSTRACT

Introduction: Allergic reaction to shunt material is rarely encountered. We report a case of hypersensitivity to silicone-based shunt material that did not subside with corticosteroid which required shunt removal.

Case presentation: A 5 years old child with spastic quadriplegic cerebral palsy was brought to our ER due to loss of consciousness. Physical examination revealed anisocoria of the pupils (2 mm / 7 mm) and Glasgow Coma Scale score of E2VxM3. Head computed tomography (CT) scan showed a fluid collection in the frontotemporal region and the appearance of schizencephaly.

Conclusion: Allergic reaction to silicone in ventriculoperitoneal shunt material is extremely rare. Polyurethane-based tubes are usually used as a substitution for silicone tubes. However, these tubes were not available and were not covered by insurance in Indonesia. Ventriculostomy is another option for selected cases.

Keywords: schizencephaly, silicone hypersensitivity, ventriculoperitoneal shunt.

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INTRODUCTION

Allergic reaction to shunt material is rarely encountered. Gower *et al.*¹ through an electron microscope found that silicone allergy might cause shunt malfunctions which could lead to breast and joint implant complications.

In 1989, Snow and Kossovsky² revealed that three patients experienced several eosinophils and giant cells due to exaggerated responses to the shunt hardware. In 1992, Goldblum *et al.*³ used ELISA to examine two patients with acute reactions to ventriculoperitoneal shunts and revealed that there were increases in serum IgG, which indicated that the two patients had immune-mediated reactions.

In this article, we aimed to report a hypersensitivity case to silicone-based shunt material that did not subside with corticosteroid which required shunt removal.

CASE PRESENTATION

A 5 years old child with spastic quadriplegic cerebral palsy was brought to our ER due to loss of consciousness. Physical examination revealed anisocoria of the pupils (2 mm / 7 mm) and Glasgow Coma Scale score of E2VxM3. Head computed tomography (CT) scan showed a fluid collection in the frontotemporal region and the appearance of schizencephaly (Fig. 1A and 1B). VP shunt procedure was performed immediately to decrease intracranial pressure. The patient experienced improvement within 2 days to GCS of E4VxM6 and was discharged 5 days postoperatively. The patient was readmitted 3 days later with redness surrounding the tube from the ear, neck, and down to the abdominal area (Fig. 2A and 2B). Laboratory results showed eosinophilia and basophilia. Corticosteroid and antibiotic were administered empirically. The patient was followed up for a week and the allergic reaction did not subside. Long-term

immunosuppression was not initiated. The patient was referred to another hospital for shunt removal and further management. The patient was followed up in two months on an outpatient basis and had no residual symptoms.

DISCUSSION

From our knowledge, this is the first case of reported hypersensitivity to silicone in patients receiving VP shunt with silicone material in Indonesia. We performed a literature search on Pubmed, EuroPMC, EBSCO, and other databases. We found several studies that reported the incidence of hypersensitivity to silicone VP shunt causing various degrees of obstruction. We propose in the future that a hypersensitivity test be performed in patients suspected of having hypersensitivity to silicone. Our suspicion of sensitivity was supported by the clinical features of the patient. The patient experienced severe itching and redness on the skin surfaces overlying the VP

Figure 1. A and B: The result of the head CT scan of the patient.

Figure 2. A and B: The skin condition of the patient.

shunt from behind the ear into the frontal thoracic/abdominal area and laboratory results revealed eosinophilia. We did not entertain the possibility of shunt infection as a possible differential diagnosis due to the fact that most infections occur in a period of approximately 6 months after shunt placement. The patient also did not show any systemic symptoms (eg. fever and leukocytosis) that support a possibility of infection.

The biomaterials used in the central nervous system are synthetic polymers, natural polymers, lipids, and silicone. They can take in many forms depending on their applications. Based on biocompatibility, there are three types of implant materials: “inert”, “tolerated”, and “bioactive”. Silicone and latex, meanwhile, are widely known

as not inert materials. Therefore, it allows the body to react differently in response to them.⁴

There are rare cases reporting silicone ventricular shunt allergies. Gower *et al.*¹ through an electron microscope, found that silicone allergy might cause shunt malfunctions which could lead to breast and joint implant complications. In 1989, Snow and Kossovsky² conducted a study on 29 patients who underwent surgery to repair ventriculoperitoneal shunts that failed to function normally. By observing their clinical and pathological findings, Snow and Kossovsky revealed that three patients experienced several eosinophils and giant cells due to exaggerated responses to the shunt hardware. In 1992, Goldblum *et al.*³ used ELISA to examine

two patients with acute reactions to ventriculoperitoneal shunts and revealed that there were increases in serum IgG, which indicated that the two patients had immune-mediated reactions. In 1994, Jimenez *et al.*⁵ revealed that there was a clinically heterogeneous entity found in three hydrocephalus patients with silicone ventriculoperitoneal shunt allergy. Although it clinically indicated a shunt-related infection, the three patients' cerebrospinal fluid (CSF) remained uncontaminated. The three patients experienced repeated skin damages over the shunt tract as well as the development of subsequent infections and fungating granulomas. To one patient, the hypersensitivity was treated by substituting the silicone shunt material with immunosuppressive polyurethane; to other patients, the shunt was taken out without a replacement. In 1999, the only case of bowel perforation caused by silicone shunt allergy was reported.⁶

Type IV hypersensitivity seems to be the most common silicone allergy. This reaction is transmitted by T cells and begins within two to seven days after the exposure.⁷ Hashimoto *et al.*⁸ reported a rare case of a patient with an abdominal CSF pseudocyst caused by silicone allergy. The patient had surgery of the meningomyelocele associated with the Chiari II malformation, and the ventriculoperitoneal shunt was installed for six months. Forty days after, the observation on abdominal CSF pseudocyst and the subsequent malfunction was conducted. It was found that the pseudocyst was caused by the increasing number of serum IgE, peripheral eosinophils, and infiltration of eosinophils in the specimen, which indicated that there was an allergic reaction. A sixth surgery to repair the ventriculoperitoneal shunt malfunction was conducted using extracted silicone. The serum IgE returned to normal after surgery and the patient was free from abdominal CSF pseudocyst for 22 months.⁸

The allergic reactions usually occur through local reactions, silicone migration, or human autoimmune disease. Emergency rooms may be required by patients if they have one of these symptoms; indolent shunt infection caused

by irritability and low-grade fever or shunt obstruction caused by aggregation of the fenestrated catheter tip with eosinophils and giant cells.⁷

Diagnosis of silicone allergy needs to be started by eliminating infection, obstruction, and other forms of shunt complications. Other potential triggers, such as intravenous or topical antibiotics like bacitracin, must also be examined.⁹ Symptom-burden patients with a high risk of latex allergy can use SPT and ImmunoCAP regularly to measure the allergen circulating them.¹⁰

Jimenez *et al.*⁵ suggested a battery of resources for assessing silicone allergies such as assays for fluorescent nuclear antibodies, human leukocyte antigen typing, immunoglobulin and complement levels, erythrocyte sedimentations rate, ribonucleic proteins, extractable nuclear antibodies, rheumatoid factor, and tissues biopsy. They also suggested performing ELISA tests for anti-silicone IgG antibodies when available. A pathological examination of the malfunctioned hardware, furthermore, is recommended.

CONCLUSION

Allergic reaction to silicone in VP shunt material is extremely rare. Polyurethane-based tubes are usually used as a substitution for silicone tubes. However, these tubes were not available and were not covered by insurance in Indonesian.

Ventriculostomy is another option for selected cases.

CONFLICT OF INTEREST

There was no conflict of interest related to the materials or methods used in this study.

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AUTHORS' CONTRIBUTIONS

Authors took part in the design of the study, contributed to data collection, participated in writing the manuscript and all agree to accept equal responsibility for the accuracy of the contents of this article.

REFERENCES

- Gower DJ, Lewis JC, Kelly Jr DL. Sterile shunt malfunction. A scanning electron microscopic perspective. *J Neurosurg.* 1984; 61(6): 1079 – 84. <https://doi.org/10.3171/jns.1984.61.6.1079>.
- Snow RB, Kossovsky N. Hypersensitivity reaction associated with sterile ventriculoperitoneal shunt malfunction. *Surg Neurol.* 1989; 31(3): 209 – 14. [https://doi.org/10.1016/0090-3019\(89\)90119-5](https://doi.org/10.1016/0090-3019(89)90119-5).
- Goldblum RM, Pelley RP, O'Donnell AA, Pyron D, Heggors JP. Antibodies to silicone elastomers and reactions to ventriculoperitoneal shunts. *Lancet.* 1992; 340(8818): 510 – 3. [https://doi.org/10.1016/0140-6736\(92\)91710-p](https://doi.org/10.1016/0140-6736(92)91710-p).
- Eymann R, Kim Y-J, Bohle RM, Antes S, Schmitt M, Menger MD, *et al.* Microstructural alterations of silicone catheters in an animal experiment: Histopathology and SEM findings. *Acta Neurochir Suppl.* 2012; 113: 87 – 90. https://doi.org/10.1007/978-3-7091-0923-6_18.
- Jimenez DE, Keating R, Goodrich JT. Silicone allergy in ventriculoperitoneal shunts. *Childs Nerv Syst.* 1994; 10(1): 59 – 63. <https://doi.org/10.1007/BF00313586>.
- Brownlee JD, Brodtky JS, Schaefer IK. Colonic perforation by ventriculoperitoneal shunt tubing: A case of suspected silicone allergy. *Surg Neurol.* 1998; 49(1): 21 – 4. [https://doi.org/10.1016/s0090-3019\(97\)00014-1](https://doi.org/10.1016/s0090-3019(97)00014-1).
- Alvis-Miranda H, Alcalá-Cerra G, Castellar-Leones SM, Escorcia HF, Moscote-Salazar LR. Latex and silicon allergy related to cerebrospinal shunt hardware: a review. *Rom Neurosurg.* 2013; 20(1): 106 – 15.
- Hashimoto M, Yokota A, Urasaki E, Tsujigami S, Shimono M. A case of abdominal CSF pseudocyst associated with silicone allergy. *Childs Nerv Syst.* 2004; 20(1): 761 – 4. <https://doi.org/10.1007/s00381-003-0904-0>.
- Hussain NS, Wang PP, James C, Carson BS, Avellino AM. Distal ventriculoperitoneal shunt failure caused by silicone allergy. Case report. *J Neurosurg.* 2005; 102(3): 536 – 9. <https://doi.org/10.3171/jns.2005.102.3.0536>.
- Kattan H, Harfi HA, Tipirneni P. Latex allergy in Saudi children with spina bifida. *Allergy.* 1999; 54(1): 70 – 3. <https://doi.org/10.1034/j.1398-9995.1999.00683-x>.

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