



# Case report: the unexpected culprit for multiple shunt revisions

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## Abstract

Silicone allergy in patients with ventricular shunts is uncommon hence easily missed. However, there are clinical features that could assist in identifying and diagnosing this condition. We discuss a case where a patient with a ventriculoperitoneal (VP) shunt presented to us with features suggestive of silicone allergy.

**Keywords** Silicone allergy · Shunt allergy · Ventriculoperitoneal shunt · Hypersensitivity

## Introduction

Silicone is a bioinert material which has been used to manufacture various implants since 1946 [1]. Following a shunt insertion, a fibrin sheath is formed around the extracranial portion of the shunt [2]. This sheath is not attached to nor does it interfere with the function of a shunt and is not considered as a hypersensitivity reaction [2]. Silicone allergy is rare and its mechanism is poorly understood. In this article, we discuss a patient who presented with silicone hypersensitivity.

## Case study

The patient was a 3-year-old boy who had a VP shunt insertion for hydrocephalus secondary to intraventricular hemorrhage at birth.

This patient initially presented with skin lumps over the shunt tract on his chest. He had multiple shunt revisions in the past for suspected infections where he experienced similar symptoms. On examination, he was crying and the skin lumps lacked signs to suggest an infection. Radiological examination

did not reveal any abnormalities. His blood leucocytes and cerebrospinal fluid (CSF) laboratory tests did not support the presence of an infection.

Nonetheless, this patient was managed with antibiotics to cover for infection and externalization of the shunt. His symptoms subsided, and he underwent a shunt revision.

Two months later, he presented twice with similar skin lesions and concurrent abdominal pain. Abdominal ultrasound showed a 2.8-cm pericatheter collection. Throughout these admissions, his laboratory tests showed no signs of infection; however, there was a presence of eosinophilia in the blood and CSF. Hence, silicone allergy was suspected, and this patient underwent a VP shunt revision with a silicone-free shunt. He has since remained symptom-free.

## Discussion

The most common complication of ventricular shunt insertions is shunt obstruction followed by infection [3, 4]. Hypersensitivity reactions to shunts are rare. Although silicone is considered as an inert biomaterial, it has been implicated to cause chronic local or systemic reactions which include inflammatory processes, foreign body reactions, and autoimmune reactions [5].

One of the earliest reports was by Gower et al. in 1984, who examined shunt tubings with electron microscopy and proposed possible hypersensitivity reactions to silicone in his patients [6]. Later in 1989, Snow and Kossovsky found that three of their patients with shunt malfunction had hypersensitivity reactions [7]. Goldblum described two cases where patients developed skin reactions and abdominal cysts around

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the catheter which were associated with elevated levels of immunoglobulin G [8]. Jimenez also encountered three patients who presented predominantly with skin breakdown [9]. Brownlee described a more complicated case where the patient had multiple skin erosions, pleural effusion after a ventriculopleural shunt insertion and colonic perforation from a VP shunt [10]. This patient also had a ventriculojugular shunt which resulted in skin erosion [10]. Hashimoto reported about a patient with recurrent abdominal distension and shunt malfunction [11]. This patient had raised peripheral eosinophils and serum immunoglobulin E, and eosinophilic infiltration of the abdominal pseudocyst wall [11].

The diagnosis of patients with silicone allergy is difficult to ascertain. However, a similar recurring pattern can be seen in patients with ventricular shunts which can help raise clinical suspicion. They develop symptoms from shunt malfunction and local reaction to the shunt such as skin erythema and erosion along the shunt tract. Shunt malfunction in these patients may be due to local inflammatory reaction around the shunt tip which results in pseudocyst formation or visceral organ perforation [10, 12].

There is yet a guideline to help establish the diagnosis of silicone allergy in patients with ventricular shunts. From the literature, authors used various methods to assist in diagnosis. Hussain stressed on excluding other causes of shunt malfunction as the initial step [12]. This may prove challenging in patients with skin breakdown or visceral organ perforation who are already at increased risk of developing concomitant infections which could mask the allergic reaction. These patients are likely to have elevated leucocytes associated with eosinophilia in the blood or CSF. Jimenez suggested performing a series of tests, measuring the erythrocyte sedimentation rate, complement (C3 and C4), immunoglobulin (IgG, IgA, IgE, and IgM), and fluorescent antinuclear antibodies (FANA) levels [9]. If FANA was positive, he proposed that extractable nuclear antibodies, ribonucleoprotein, rheumatoid factor, tissue biopsy, and human leukocyte antigen typing should be performed when possible. Histology of shunt tracts or shunt tips may demonstrate the presence of giant cells, inflammatory cells, and foreign body granulomas [9]. Specimens from an abdominal pseudocyst wall can also demonstrate similar findings where the wall has been infiltrated by eosinophils [11].

Jimenez treated one of his patients with a short course of cyclosporin and up to 1 year of prednisolone. In a patient with abdominal pseudocyst, Hashimoto performed a shunt revision by inserting the peritoneal catheter under direct vision laparoscopically [11]. Most patients with suspected silicone allergy underwent a shunt revision using a silicone-free shunt, as in our patient, which helped to resolve their symptoms. We did not treat our patient with immunosuppressants or steroids as we did not see the need to do so. We performed a shunt revision on the side contralateral to his most recent shunt without laparoscopic guidance.

## Conclusion

Although the occurrence of silicone allergy in patients with ventricular shunts is rare, early diagnosis helps to save patients from multiple admissions, shunt revisions and risks of shunt infections, and obstructions. This diagnosis should be suspected in patients with cutaneous manifestations, shunt malfunction, and eosinophilia. Initial investigations should include ruling out infections, mechanical causes for shunt failure and rheumatological conditions. CSF sample and shunt tip also should be sent for cell count and culture studies. The treatment of these patients should be as any other allergy management where the precipitating agent is removed.

## Compliance with ethical standards

**Informed consent** Informed consent was obtained from all individual participants included in the report.

**Conflict of interest** On behalf of all authors, the corresponding author states that there is no conflict of interest.

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