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ORIGINAL ARTICLE



Silicone allergy mimicking shunt infection

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ABSTRACT

Purpose: The purpose of this case report is to highlight silicone allergy as a rare cause of ventriculo-peritoneal shunt complication, often mimicking infection.

Materials and methods: Information regarding the patient presented was obtained by review of their medical notes, both paper and electronically. A literature review was performed by searching Pubmed using the following terms; silicone allergy and shunt allergy, to identify similar publications.

Results: Nine publications between 1994 and 2019 formed the basis of the literature review. The primary aim was to identify clinical presentations that may help to identify silicone allergy as the cause of shunt complication.

Conclusion: Silicone allergy is a rare cause of ventriculo-peritoneal (V-P) shunt failure and can mimic infection. This diagnosis should be considered with a predominantly lymphocytic CSF leucocytosis and no bacteriological evidence of a responsible organism, a peripheral eosinophilia and an intense fibrosis around a recently placed V-P shunt, as described in this case report.

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KEYWORDS

Silicone allergy;
hydrocephalus; ventriculo-peritoneal shunt;
hypersensitivity

Case report

We describe an 18 year old with a diagnosis of benign external hydrocephalus treated by ventriculo-peritoneal (V-P) shunting in childhood. She had a number of revisions, notably one for an abdominal pseudocyst, which resulted in placement of a ventriculo-atrial (V-A) shunt. At this presentation, her shunt had not been revised for five years until she sustained trauma to the shunt in the parietal region, resulting in wound breakdown. A new bactiseal V-P shunt was placed but she subsequently had a number of episodes of headache, neck and abdominal pain and vomiting. On inspection there was a marked fibrotic reaction in the skin along the course of the shunt. Four months after this shunt insertion there was an area of wound dehiscence over an incision on the chest wall which prompted a shunt exploration.

The ratio of cerebro-spinal fluid (CSF) white cell count (WCC) to red cell count (RCC) was elevated with a CSF WCC of 9/uL and a CSF RCC less than 1/uL. The shunt was removed and empirical antibiotics prescribed given the clinical suspicion of infection. The CSF WCC returned to normal and a new V-P shunt was again placed. She then developed a rapid meningitic response within twelve hours of the operation and CSF sampling revealed a WCC of 291/uL, 70% of which were lymphocytes, with a RCC of 638/uL. CSF glucose and protein were normal.

The CSF leucocytosis did not normalise with removal of the shunt and replacement with a bactiseal external ventricular drain, a two-week course of empirical intravenous antibiotics and subsequent intrathecal antibiotics. Her peripheral blood samples revealed an eosinophilia at this time and no causative organisms were identified on CSF and peripheral microscopy and cultures.

This led to the diagnosis of a silicone allergy. We discussed the case with the Immunology team who advised that there was

no specific testing that could confirm this, however serum IgE and silicone antibody testing was considered.

Approval for a silicone-free shunt was required from the Medicines & Healthcare products Regulatory Agency (MHRA) as a humanitarian request as polyurethane shunts do not have a CE mark.¹ This resulted in a six-week delay where the patient had external ventricular drainage prior to successful insertion without complication or subsequent meningitis. The patient has been well with no further admissions secondary to a shunt complication.

Discussion

Upon review of cases of suspected silicon shunt allergy in the literature, the pathophysiology and criteria for diagnosis are unclear. There are few reports of ventriculo-peritoneal shunt silicone allergy in the literature, but this is likely an under-recognised cause of shunt complications due to the ambiguous clinical presentation.

Nine published case reports published between 1994 and 2019 described patients with suspected shunt allergy leading to shunt malfunction, either obstruction or infection.²⁻⁹ Some had described either positive blood or CSF cultures, which were attributed to superimposed bacterial infection.^{3,5} Some serious consequences associated with V-P shunt silicon allergy have also been published, such as bowel perforation due to erosion by the shunt tubing.⁵

The patients present with symptoms commonly associated with malfunction such as headache, fever, malaise and some presented with abdominal pain or abdominal pseudocyst formation. Seven of the cases describe skin changes along the tubing site on examination, commonly granulomatous tissue formation and erosion along the insertion site. Obstruction of the distal catheter with eosinophils and giant cells accumulating at the catheter tip

may occur.^{2,6} Pathologically, patients have been noted to have eosinophils and giant cells in tissue enveloping the shunt hardware.¹⁰ The mechanism of silicone allergy seems to be a type IV or delayed hypersensitivity reaction.¹¹

Most of the case reports detailed the investigations undertaken, to include; complete blood count, c-reactive protein, peripheral blood cultures and CSF cultures. Both peripheral and CSF eosinophilia appear in five cases, but negative inflammatory markers and cultures led to the diagnosis of silicone allergy in several patients.^{5,7–9}

The CSF may not show overt signs of infection; however, the presence of eosinophilia will help aid the suspected diagnosis which if made early, will reduce the likelihood of repeated hospital admissions, and the need for repeated shunt revisions.⁷ This is of particular importance when considering the costs associated with multiple shunt revisions, as outlined by Patwardhan *et al.* in their review of the costs of shunt revision in the USA.¹²

Treatment of the silicone allergy was generally with the placement of a silicone-free shunt, with one patient proceeding to have a ventriculostomy and one patient remaining well without placement of any further shunts.³ Four cases described the use of polyurethane shunts but did not detail their production, while two cases described extracted silicone tubes and one simply a 'silicone-free shunt'.^{3–7}

Silicone elastomers have been presumed to be immunologically inert but hypersensitivity reactions to silicon have been noted in other fields. In otorhinolaryngology there have been reported reactions to cochlear implants and tracheal tubes following laryngectomy,¹³ however both are rare,¹¹ and similar reactions have been observed with silicone breast implants.¹⁴

The silicone-free polyurethane shunts are felt to be comparable to standard silicone shunt with regard to both their function and resistance to malfunction as the products have similar properties. It is felt that the addition of immunosuppressive therapy is not required when placing a polyurethane shunt.⁶ The shunt used in this case was produced in Dublin, Ireland, by Medtronic on request. It comprises a delta valve, which is a fixed pressure valve with an anti-siphon mechanism.

Conclusion

This case report and literature review highlights a rare complication of V-P shunting which can mimic infection or obstruction. The features of a predominantly lymphocytic CSF pleocytosis,

lack of organisms seen on CSF microscopy or culture, a peripheral eosinophilia and marked tissue reaction to the silicone shunt, should help make the diagnosis earlier.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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