Cardiology in the Young

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Cite this article: Othman T and Eldadah O (2022) Treatment of chronic resistant paediatric idiopathic pericardial effusion with intrapericardial injection of corticosteroids 4-year-old girl with rapid full resolution after intrapericardial injection of corticosteroids. *Cardiology in the Young* **32**: 1169–1171. doi: 10.1017/S1047951121004637

Received: 8 September 2021
Revised: 6 October 2021
Accepted: 31 October 2021
First published online: 15 December 2021

Keywords:
Pediatric; pericardial effusion; intrapericardial steroids

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Treatment of chronic resistant paediatric idiopathic pericardial effusion with intrapericardial injection of corticosteroids 4-year-old girl with rapid full resolution after intrapericardial injection of corticosteroids

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Abstract

Non-steroidal anti-inflammatory drugs are the conventional treatment for pericarditis. However, some patients will still suffer from persistence pericardial effusion despite exhausting all conventional management options. A 4-year-old girl with idiopathic pericardial effusion who did not respond to 2 months of conventional therapy had complete resolution of effusion within 5 days, with no recurrence after administration of intrapericardial steroids. As far as we know, this is the first published paediatric case who has shown a similar outcome to that seen in adult studies.

Pericarditis is the most common disease of the pericardium and accounts for 5% of all children who present with chest pain, with a risk of recurrence of about 30% within 18 months from presentation.[1,2]

The 2015 European Society of Cardiology Guidelines for the diagnosis and management of pericardial diseases recommends the following medications for treatment of pericarditis in children:

High-dose non-steroidal anti-inflammatory drugs, given until complete symptoms resolution, are the first-line therapy, while colchicine or anti-interleukins-1 drugs are to be given as an adjunct therapy for recurrent pericarditis. Systemic corticosteroids were not recommended due to the severity of side effects, but if given, then the minimal effective dose should be used.[3]

Data review

There are currently no randomised controlled trials in children with pericardial diseases, with all medical therapies being off-label. Most of our experience in managing these patients is adopted from adult studies. While the use of non-steroidal anti-inflammatory treatment or colchicine was proven to be effective,[4] some patients still suffer from persistent prolonged effusion despite the long course of medication.

In our current case, we were faced with this dilemma after exhausting all conventional management options.

Reviewing the literature, we found a growing evidence of the efficacy of intrapericardial steroids administration.

The first case of cardiac tamponade successfully treated by pericardiocentesis and intrapericardial injection of methylprednisolone was described by Sharf et al in 1976.[5]

Maisch B and college found that in this type of patient, the use of intrapericardial steroids could be an effective treatment with minimal systemic side effects and long-term efficacy with no recurrence. The direct intrapericardial application of drugs has the advantage of giving a high local dose with little systemic side effects.[6]

Another study by the same group evaluated the efficacy and safety of intrapericardial treatment with the crystalloid corticosteroid triamcinolone in autoreactive pericardial effusion; it found that it resulted in symptomatic improvement and prevented recurrence in 92.6% of patients after 3 months and in 86.0% after 1 year.[7]

Case description

A 43-month-old girl who had previously been healthy, presented with a history of poor activity, mild chest discomfort, face puffiness, and bilateral foot swelling of 2 months duration.

Table 1. List of lab work up done which was all unremarkable supporting the diagnosis of idiopathic pericardial effusion.

Investigations	Results
Respiratory viral multiplex	Negative
Pericardial fluid culture	Negative
Pericardial fluid analysis	Unremarkable
Hepatitis C,B	Negative and immune
Bacterial multiplex	Negative
Brucella titre	Negative
Parvovirus PCR	Negative
EBV (PCR)	Negative
TB quantiferon	Negative
Thyroid function tests	Normal
CBC, ESR, CRP	Normal
ANA	Negative
Anti-ds DNA	Negative
Anti SSA, anti SSB, anti-SM	Negative
Complement C3, C4	Normal
Total compliment activity	Normal
Pericardial tissue pathology	No significant abnormality
Pericardial fluid cytology	Negative for malignant cells

The symptoms were preceded by mild upper respiratory tract infection, but there was no history of fever. Her mother is a known case of systemic lupus erythmatosis.

The first echo performed at presentation showed a large pericardial effusion with impending tamponade and normal intracardiac anatomy. Emergency pericardiocenthesis was done and 280 ml (19/ml/kg) of pericardial fluid was removed.

Fluid analysis was unremarkable. Serum albumin was normal, and urine was negative for protein so nephrotic syndrome spectrum was excluded. Rheumatology, immunology, and infectious disease teams requested a full work up which was all unremarkable (Table 1).

Our impression was that her presentation was most likely explained by post infectious inflammatory pericarditis or idiopathic pericardial effusion.

She was started on furosemide, methylprednisolone for 5 days, and then aspirin with Colchicine (Fig 1).

However, despite all medical management, she continued to drain from pericardial tube with fluctuating amounts of 2–10 ml/kg/day. Serial echoes showed moderate to minimal pericardial effusion depending on the drain amount with no significant improvement

After multiple failed attempts to clamp the drain with significant re-accumulation of fluid within 24 hours from clamping, she was resumed back on prednisolone and aspirin was stopped.

Following 20 days in hospital with no progress, the creation pericardio-pleural window was carried out after careful multidisciplinary discussion. A sample of the pericardium was taken for pathology assessment and showed no abnormality.

However, the drain persisted in having a high output (around 10 ml/kg/day) and so another set of rheumatology work up tests was requested which later came back normal. A trial of Indomethacin for 6 days was attempted with no improvement, so it was discontinued. Finally, a therapeutic trial of Anakinra was given for 5 days without improvement. The final impression was that it was unlikely the patient's condition was related to a rheumatological cause.

At this point, the patient had been in hospital for 2 months with no response to all medical management; the case was re-discussed and after reviewing the published data mentioned above the team agreed to a trial of insertion of local steroids through tubing directly into the pericardial space.

All medications were stopped, and drains were clamped a day before the procedure. The procedure was performed in the cath lab under transthoracic echocardiography guidance with conscious sedation. An injection of 30 mg (2 mg/kg) of Methylprednisolone through the pleural and pericardial tubes each was given and pericardial and pleural drains kept clamped for 24 hours then unclamped. She was started on oral prednisolone with a dose of 2 mg/kg/day.

The fluid amount in the drains was observed closely in the following days, and it became less than 3 ml/kg/day within 5 days. The drains were clamped on day 5 after the procedure. Echos 24 hours and 48 hours post clamping showed minimal pericardial

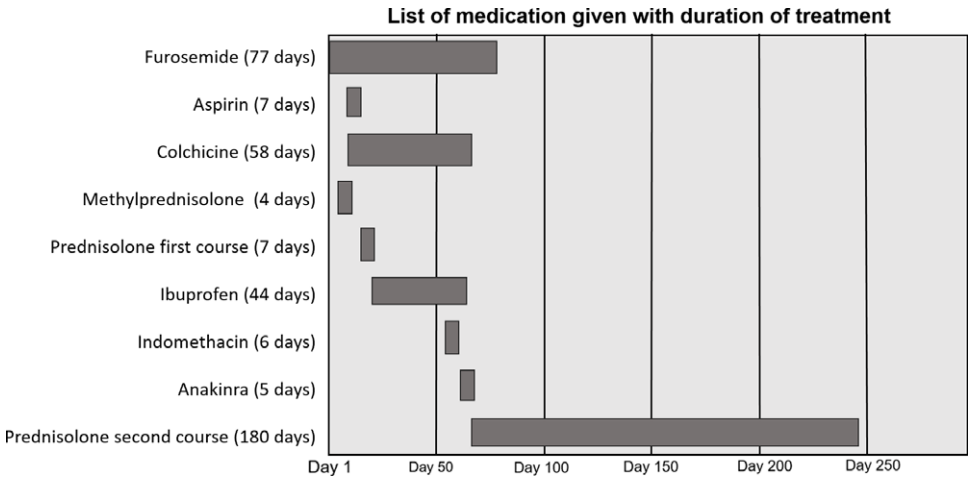

Fig. 1. Bar chart showing the time line and total duration of medication given during admission and after discharge (discharged on day 78).

https://doi.org/10.1017/S1047951121004637 Published online by Cambridge University Press

effusion. They were removed on day 7, and Echo follow-up on day 10 showed no pericardial effusion. The girl was finally discharged on oral prednisolone with OPD follow-up in 1 week which also showed no pericardial effusion.

Follow-up at the 1 and 3 months marks was also normal, and prednisolone was weaned off gradually over 3 months. She remained medically free after 2 years with no more effusion.

As described above, the use of intrapericardial steroids was in this case a very effective treatment with no recurrence or major complications. As far as we know, this is the first published case to study this method in paediatric idiopathic pericardial effusion which showed a similar outcome to adult studies. However, more studies with larger number of patients are needed to confirm our experience.

Acknowledgements. We would like to acknowledge Dr Omer Altamimi for his kind effort and input in managing this patient.

Financial support. This case report received no specific grant from any funding agency, commercial, or not-for-profit sectors.

Conflicts of interest. None.

Ethical standards. Patient confidentiality was maintained at all level, and this case report was approved by institutional review board in national guard hospital with approval no RC 20\602\R.

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https://doi.org/10.1017/S1047951121004637 Published online by Cambridge University Press