

# A Rare Complication After Surgical Repair of Atrioventricular Septal Defect: Chylopericardium: Case Report

## Atriyoventriküler Septal Defekt Ameliyatı Sonrası Nadir Görülen Bir Komplikasyon: Şiloperikardiyum

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**ABSTRACT** A four months old male infant with Down Syndrome underwent cardiac surgery for repair of atrioventricular septal defect. An control echocardiogram performed on 16th day of the operation, revealed pericardial effusion and the patient was hospitalized for treatment. Pericardiocentesis was performed under the guidance of echocardiography. The fluid obtained appeared chylous. The blood triglyceride level was 647 mg/dL, total protein level was 4,5 g/dL. Biochemical evaluation of the fluid revealed that the triglyceride level was 1253 mg/dL, protein: 4.3 g/dL, thus firstly we performed diet therapy and thereafter tube pericardiostomy. After 5 days of tube drainage the pericardial effusion resolved and there was no recurrence in follow-up.

**Key Words:** Cardiac tamponade; pericardial effusion; pericardiocentesis

**ÖZET** Down sendromu bulunan 4 aylık erkek çocuğa atriyoventriküler septal defekt tanısı ile düzeltme ameliyatı yapıldı. Ameliyattan sonraki 16. günde kontrol amaçlı yapılan ekokardiyografi ile perikardiyal efüzyon saptandı ve hasta tedavi amacı ile tekrar hastaneye yatırıldı. Ekokardiyografi eşliğinde perikardiyosentez yapıldı. Elde edilen sıvının şilöz özellikte olduğu görüldü. Serumda trigliserit düzeyi 647 mg/dL, total protein seviyesi ise 4,5 g/dL iken sıvının biyokimyasal analizinde trigliserit seviyesinin 1253 mg/dL, protein seviyesinin ise 4,3 g/dL olduğu saptandı. Şiloperikardiyum düşünülerek önce diyet tedavisi, bu tedavi ile efüzyonda gerileme olmayınca daha sonra tüp perikardiyostomi uygulandı. Tüp drenajından 5 gün sonra perikardiyal efüzyonun gerilediği ve izlemde şiloperikardiyumun tekrarlamadığı görüldü.

**Anahtar Kelimeler:** Kardiyak tamponad; perikardiyal efüzyon; perikardiyosentez

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Chylopericardium is a rather rare situation usually observed after cardiac surgical procedures. It's not that common after intrapericardial procedures as thoracic duct is not close to the surgical dissection zone. Chylopericardium can be observed due to increase of lymphatic permeability in children with chromosomal anomaly and lymphatic dysplasia. It can also be seen after cardiac surgical procedures leading to trombus formation in large venous vessels or damage to lymphatic vessels of thymus. Lymphatic tissue of thymus is larger in children and thus it's more common among them. Here we report a case with chylopericardium after congenital cardiac surgery, a rare situation with potential important problems.

## CASE REPORT

A four months old male baby with Down Syndrome underwent cardiac surgery for repair of atrioventricular septal defect (AVSD). The postoperative course was uneventful and he was discharged two weeks after the operation. An echocardiogram performed on 16<sup>th</sup> day of the operation, revealed pericardial effusion and the patient was hospitalized. He was in good condition and had a rhythmic and strong pulsation. He had a grade 3/6 systolic murmur on the left of sternum. Hepatomegaly was not observed. Laboratory findings were; Hb:10.1 g/dL, Hct: 31.5%, platelet count: 189 000/mm<sup>3</sup>, C reactive protein:5.9 g/dL. Triglyceride level was 647 mg/dL and total protein was 4.5 g/dL. Cardiomegaly was present on telecardiography. Electrocardiogram showed a QRS wave with low amplitude. Echocardiogram revealed massive pericardial effusion in the posterior area of the left ventricle and the basal region. Pericardiocentesis was performed under the guidance of echocardiography. The fluid obtained appeared chylous. Biochemical evaluation of the fluid revealed that the triglyceride level was 1253 mg/dL, protein: 4.3 g/dL and white cell count: 16 300 /mm<sup>3</sup>. No microorganisms were seen in microscopic evaluation. Wright smear showed 20% lymphocytes and 80% erythrocytes. Modification of the diet with medium chain fatty acids was ineffective and thus tube pericardiostomy was performed. After 5 days of tube drainage the effusion resolved and there was no recurrence in follow-up.

## DISCUSSION

Chylopericardium is a rather rare situation which can be observed after cardiac surgical procedures, trauma and due to occlusion of drainage of thoracic duct to subclavian vein.<sup>1</sup> It can rarely be observed after tuberculosis, neoplasm or congenital lymphangiomatosis.<sup>2</sup>

While pericardial effusions can be observed in almost 30% of all cardiac surgical procedures, chylopericardium is observed in less than only 0.22% of them. It is mostly observed on the 10<sup>th</sup> postoperative day and 1% can end up with tamponade.<sup>3,4</sup> In a study by Kan et al. which was performed between

1988 and 2004, the rate of chylopericardium was 0.12% after intrapericardial surgical procedures.<sup>5</sup>

The pathogenesis of chylopericardium arising after surgery is not clear. Still in some cases with chromosomal anomalies such as Down Syndrome, Noonan Syndrome and Turner Syndrome, the increase of lymphatic permeability is known to cause chylopericardium as in our case.<sup>6,7</sup>

The main characteristic of the chylous effusion is the existence of a milky fluid within pericardium. In 2005, Densupsoontorn et al. studied 16 cases, 14 of whom had chylothorax and 2 had chylopericardium, and found that the protein level of the fluid was 2.4-7.4 mg/dL, the triglyceride level was 59-1684 mg/dL and higher than the plasma triglyceride level.<sup>8</sup> In our case, the triglyceride level of the fluid was 1253 mg/dL, plasma triglyceride level was 647 mg/dL and the protein level of the fluid was 4.3 g/dL. These results were parallel with Densupsoontorn's study.

In the same study the mean period between the surgical procedure and the time of diagnosis was 13 (range 3-30) days. The mean period of the tube drainage was 12.1 (range 3-29) days.<sup>8</sup> In our case the drainage was initiated on 16<sup>th</sup> postoperative day and it lasted for 5 days.

Chylopericardium is a rare situation observed after cardiac surgical procedures. In 4 of 16 cases reported by Campbell et al.<sup>6</sup> and in one of 4 cases presented by Chung-Dan Kan chylopericardium was observed after repair of atrioventricular septal defect and all cases had Down Syndrome. In our case chylopericardium was also observed after surgical repair of atrioventricular septal defect in a patient with Down Syndrome.

The initial treatment of chylopericardium is administration of a diet with medium chain fatty acids.<sup>9</sup> Total parenteral nutrition (TPN) is rarely indicated. Campbell et al. reported successful results by initiating diets with short and medium chain triglycerides (MCT) along with percutaneous pericardial drainage in most of their 16 cases.<sup>6</sup>

In Densupsoontorn's study MCT and TPN diets were administrated for 29.8 (14-47) days. We

**TABLE 1:** Clinical characteristics of patients with chylopericardium and AVSD.

	Diagnosis	Age	Gender	Syndrome	Time of diagnosis	Treatment
Campbell MR et al.	AVSD	3 months	Female	Down	29 <sup>th</sup> day	Diet
	AVSD	4 months	Female	Down	19 <sup>th</sup> day	Diet
	AVSD	5 months	Female	Down	13 <sup>th</sup> day	Diet
	AVSD	5 months	Female	Down	6 <sup>th</sup> day	
Kan C-D et al.	AVSD	12 months	Male	Down	10 <sup>th</sup> day	Diet
Gursu HA et al.	AVSD	4 months	Male	Down	16 <sup>th</sup> day	Diet+tube pericardiostomy

AVSD: Atrioventricular septal defect.

have used MCT diet alone for 6 days. Nguyen et al. reported a success rate of 84% in their 25 patients, 22 of whom had chylothorax and 3 had chylopericardium, by administrating MCT and/or TPN without additional surgical procedures.<sup>10</sup>

Pericardiocentesis under guidance of echocardiography and placement of pigtail catheter by percutaneous pericardial approach are safe and effective treatment options.<sup>11,12</sup> Campbell et al. performed pericardial drainage for 12 of their 16 cases with chylopericardium. In his series there were 4 patients with AVSD and pericardial drainage was not required for any of these patients. Pericardiocentesis under echocardiography was performed initially in our case who also had AVSD. Despite dietary treatment, effusion did not resolve and tube pericardiostomy was made. Somatostatine was rarely used for cases with chylopericardium.<sup>6,13</sup> We also did not use somatostatine for our case.

A thorough literature review revealed that there are 20 cases with chylopericardium arising after intrapericardial surgery including our case (Table 1). Six of these patients had undergone surgery for AVSD, two for AVSD-tetralogy of Fallot, four for atrial septal defect, three for tetralogy of Fallot, two for transposition of great arteries and one each for aortic stenosis, pulmonary stenosis and ventricular septal defect. Ten of these patients had chromosomal anomalies, 9 of whom had Down Syndrome. All patients with AVSD had Down Syndrome.

In conclusion we wanted to remind the risk of chylopericardium after cardiac surgical procedures, especially in patients with AVSD and Down syndrome.

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