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CASE REPORT / OLGU SUNUMU

Ostium secundum atrial septal defect-related post-partum death of an adult: An autopsy case

Ostium sekundum tip atrial septal defekt'e bağli doğum sonrasi ölüm: bir otopsi olgusu

Eyyüp Yılmaz[1], Gökhan Ersoy[2], S.Serhat Gürpınar[3], Sedat Tavşanoğlu[4]

ABSRACT

Atrial Septal Defect (ASD) is the most common type of congenital heart disease in adolescents and adults with a frequency of 10-15 %. Ostium secundum type ASD has a multifactorial heredity pattern and is almost always sporadic.

Herein, we presented a maternal mortality case of ASD that died after nine days after delivery. She applied to the hospital three times with non-specific symptoms but died without an accurate diagnosis. At autopsy, there was evidence of episiotomy in recent delivery in genital region. The weight of the heart was 380 grams, hypertrophy of papillary muscles and an ostium secundum type ASD with 1,6 x 1,1 cm dimensions were detected. Microscopic examination of heart sections revealed hypertrophy, hyperemia, and focal, minimal colliquative myocytolysis. Electrocardiogram findings and autopsy findings were evaluated together and in conclusion, our opinion on the cause of death was circulatory disturbance resulting from chronic heart disease.

Because of this, congenital heart disease which is a rare pathology and a cause of death in adult population, we presented the findings of this case.

Key words: Congenital heart disease, atrial septal defect, ostium secundum, maternal mortality, autopsy.

ÖZET

Atrial Septal Defekt (ASD) erişkin ve genç ergenlerde % 10-15 sıklık oranı ile en yaygın görülen konjenital (doğuştan) kalp hastalığıdır. Ostium Sekundum tipi ASD, multifaktöryel heredite (kalıtım) ile ilgili olup hemen hemen daima sporadiktir.

Burada, ölümünden 9 gün önce doğum yapmış ASD'li bir anne ölümü olgusu sunmaktayız. Doğumdan 3-4 gün sonra kendisini iyi hissetmemesi üzerine toplam üç kez, belirgin olmayan tıbbi yakınmaları ile hastaneye başvurmuş ancak doğru tanı konulamadan ölmüştür. Yapılan otopsisinde, genital bölgede doğum esnasında uygulanmış epizyotomi bulgusu saptandı. Kalp ağırlığı 380 gram olarak tartıldı. Papiller kasların hipertrofik görünümde oldukları ve atrial septumda 1,6x1,1 cm'lik ebatta ölçülen ostium sekundum tipi ASD olduğu izlendi. Histopatolojik incelemede; kalpte hipertrofi, hiperemi, fokal minimal kollikuatif myositoliz tespit edildi. Ölüm öncesi tıbbi yakınması, çekilen elektrokardiyogram ve otopsi bulgularına göre; kişinin ölümünün kendinde mevcut kronik kalp hastalığına bağlı gelişen solunum ve dolaşım yetmezliğinden ileri geldiğine karar verildi.

Literatürde yetişkin dönemde gerçekleşmiş ölüm olgularının çok az olması ve ender rastlanılması, özellikle olgumuza ölüm sonrası tanı konulması nedeniyle olgu sunulmaya değer bulunmuştur.

Anahtar kelimeler: Konjenital kalp hastalığı, atrial septal defekt, ostium sekundum, anne ölümü, otopsi.

INTRODUCTION

Atrial septal defect (ASD) is the most common type of congenital heart disease in adolescents and adults with a frequency of 10-15%.[1] Among these, Ostium secundum type ASD has a multifactorial heredity pattern and it's almost always sporadic.[2] The anomaly is inherited as an autosomal dominant trait.[3]

The mortality is about 3% in patients with pulmonary vascular disease and the most common causes of mortality are chronic cardiac insufficiency, pulmonary and systemic emboli (especially

[1] Directorship of Forensic Medicine Branch, Diyarbakır, Türkiye,
[2] University of Istanbul, Institute of Forensic Medicine, İstanbul, Türkiye,
[3] Directorship of Forensic Medicine Branch, Gaziosmanpaşa, İstanbul, Türkiye,
[4] University of Istanbul, Cerrahpaşa Medical Faculty, Department of Cardiology, İstanbul, Türkiye

***Yazışma Adresi /Correspondence:** Dr. Eyyüp Yılmaz,*
Directorship of Forensic Medicine Branch, Diyarbakır/ Türkiye Email: eypyilmaz@hotmail.com

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cerebral), arrhythmias, (such as, supraventricular tachycardia) bronhcopulmonary infections and hypertension. The onset of significant symptoms is seen in the fourth decade. The principal age of death is around fifty years in medically treated patients.[1]

CASE REPORT

Our case was a primigravida and primipara female and was 32 years old, who has admitted three times to different hospitals between postpartum 7th and 9th days. Physicians thought the diagnosis was postpartum infection but the patient has been discharged with only an anti-inflammatory prescription on first appeal. The day after, she received antibiotic medication at the second hospital and has been discharged again. Finally, on the postpartum 9th day, she presented to the third hospital with severe dyspnea and died there.

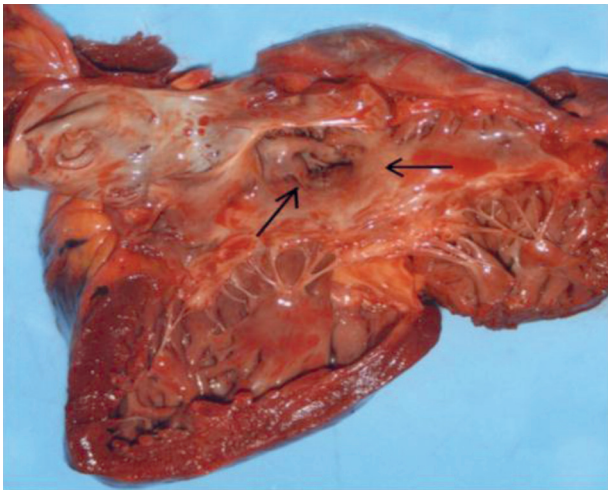

Figure 1. A huge septal defect can be seen at the center of upper half of picture

Autopsy findings

The body was measured 165 cm. height and weighted 70 kg. at autopsy. There was evidence of applied episiotomy in recent delivery in genital region. The weight of the heart was 380 grams. Macroscopically, hypertrophy of papillary muscles and an ASD of ostium secundum type measuring 1, 6 x 1, 1 cm. were detected. Thickness of the right ventricle was measured 1cm. (Figure 1). Microscopic examination of heart sections revealed hypertrophy, hyperemia, and focal, minimal colliquative myocytolysis. Examination of the lung revealed pulmonary hypertensive changes like medial thickening and plexiform structure. There were no thrombotic plugs and emboli findings in the cardiovascular system in macroscopic and microscopic examination.

Medical history

There was no medical information about the pregnancy period or manner of delivery. There was an electrocardiography (ECG) sheet which was recorded at the second hospital one day before death and P-pulmonale, right axial deviation, right bundle branch block were detected by the board cardiologist.

Board decision (Cause of death)

The complaints of the patient 2-3 days prior to death, ECG findings and autopsy findings were evaluated together. We concluded that the cause of death was circulatory disturbance resulting from chronic heart disease. Chronic cardiac illness was accepted as the main cause of death by the First Specialty Board of the Council of Forensic Medicine.

DISCUSSION

During pregnancy, hormone mediated increase in blood volume and heart rate, decreased systemic vascular resistance and arterial pressure are caused by a significant increase in cardiac output.[4]

In cases related with pulmonary hypertension, findings related to signs and symptoms begun to develop in the early twenties. Especially during pregnancy, its progress has also been rapid.

The maternal mortality varies between 27% and %66. If pregnancy occurs, abortion is the main strategy in management. If the patient decides to continue pregnancy, the patient should be hospitalized from 20 weeks until delivery.[5]

More cases with pulmonary embolism are reported and most of them mentioned a strong association with maternal death.[5-6]

Eisenmenger's syndrome in pregnancy is a rare and serious cardiac condition.[6] There is an agreement in the literature that pregnancy is poorly tolerated in Eisenmenger's Syndrome.[7]

Right ventricular hypertrophy, existence of a defect in atrial septum and pulmonary hypertensive vascular changes are the main pathologic changes

found at autopsy for this kind of cases. Our investigation exhibited all of these findings. Beside this, there were supportive electrocardiographic alterations. Although we had no information about the medical history about the deceased, all these findings can be used as strong evidences about the cause of patient's collapse.

On the other hand the delay in diagnosis may also remind medical malpractice in this case. Although she is admitted to the hospital three times, inability to find an accurate diagnosis is thought to be provoking.

This case is important to us for the following reasons

• The case is demonstrative for the probability of ASD patients that can be remained without diagnosis until adult age.

• The case bears most of the characteristic pathologic findings for ASD and Eisenmenger's Syndrome.

• The deceased had completed the pregnancy period and delivered despite the fact that she had not taken any medical support and didn't have any proper diagnosis.

In conclusion, it can be suggested to clinicians that risk level of each patient should be determined with a thorough examination before pregnancy and pregnancy should also be guided accordingly.

Note

This case report was submitted as a poster presentation in 1st International Forensic Sciences Congress, 8-11 October 2008, Istanbul, Turkey.

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