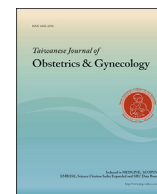




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## Case Report

# Late-onset fetal bilateral pleural effusions associated with Down syndrome

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## ABSTRACT

**Objective:** We present two cases of late-onset bilateral fetal pleural effusions associated with fetal Down syndrome.

**Case reports:** **Case 1.** A 33-year-old Vietnamese woman had undergone regular sonographic examinations since 23 weeks of gestation and no abnormality had been noted. However, bilateral moderate pleural effusions were found at 33 weeks of gestation, and massive pleural effusion, ascites and polyhydramnios developed at 34 weeks of gestation. Aspiration of the pleural effusion was subsequently performed. Clinical laboratory surveys of the aspiration fluid excluded toxoplasmosis and cytomegalovirus infection. Cytogenetic analysis of cultured lymphocytes derived from pleural effusion revealed a karyotype of 47,XX,+21. The parents elected to continue the pregnancy. Intrauterine fetal demise occurred at 37 weeks of gestation, and a macerated female baby was delivered. Postnatal cytogenetic analysis of the umbilical cord confirmed the prenatal diagnosis.

**Case 2.** A 41-year-old Pakistani woman had undergone regular sonographic examinations and no abnormality had been noted. However, isolated bilateral mild pleural effusions were noted at 27 weeks of gestation. Amniocentesis revealed a karyotype of 47,XY,+21 and simultaneous array comparative genomic hybridization analysis of uncultured amniocytes confirmed the diagnosis of Down syndrome. The pregnancy was subsequently terminated.

**Conclusion:** Fetuses with Down syndrome may present late-onset bilateral pleural effusions. Prenatal diagnosis of late-onset bilateral pleural effusions should raise the possibility of fetal Down syndrome and cytogenetic investigation is warranted.

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## Introduction

Pleural effusion is defined as fluid accumulation in the pleural space and has been estimated to occur in 1:10000–1:15000 pregnancies [1–3]. However, Hashimoto et al. [4] in a prospective study reported a higher incidence of 1.2% in low-risk obstetric cases at

7–10 weeks of gestation. Fetal pleural effusion can be classified as primary and secondary pleural effusion. Primary fetal pleural effusion is caused by lymphatic vessel anomalies or thoracic cavity defects [5,6]. Secondary fetal pleural effusion includes immune hydrops such as Rh or ABO incompatibility, and non-immune hydrops such as chromosomal abnormalities, congenital cardiac anomalies, genetic disorders, infections, metabolic diseases, and hematologic diseases [6,7].

Here we present two cases of late-onset secondary pleural effusion associated with Down syndrome.

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## Case reports

### Case 1

A 33-year-old Vietnamese multigravid woman had undergone regular sonographic examinations since 23 weeks of gestation and no sonographic abnormality had been noted. She had not received any Down syndrome screening because of her religious belief. However, bilateral moderate pleural effusions were found at 33 weeks of gestation, and massive pleural effusion, ascites and polyhydramnios developed at 34 weeks of gestation (Figs. 1 and 2). A fetal therapy of thoracentesis was suggested and an aspiration of 80 cc clear yellowish pleural fluid was subsequently made. Clinical laboratory surveys of the aspiration fluid excluded toxoplasmosis and cytomegalovirus infection. Cytogenetic analysis of cultured lymphocytes derived from aspirated pleural fluid revealed a karyotype of 47,XX,+21. The parents elected to continue the pregnancy. Unfortunately, intrauterine fetal demise occurred at 37 weeks of gestation, and a macerated 3202 g female baby was delivered. Postnatal cytogenetic analysis of the umbilical cord confirmed the prenatal diagnosis.

### Case 2

A 41-year-old Pakistani multigravid woman had undergone regular sonographic examinations and no abnormality had been noted. She refused any prenatal non-invasive or invasive diagnosis of Down syndrome because of her religious belief. However, mild isolated bilateral pleural effusions were noted at 27 weeks of gestation. The risk of chromosome abnormalities was explained to the parents and amniocentesis was suggested. Amniocentesis revealed a karyotype of 47,XY,+21 and simultaneous array comparative genomic hybridization analysis of uncultured amniocytes confirmed a diagnosis of Down syndrome. The pregnancy was subsequently terminated, and a 1900 g male fetus was delivered.

## Discussion

The particular aspect of this presentation is the late-onset occurrence of bilateral pleural effusions associated with fetal Down syndrome. Despite the introduction of Down syndrome

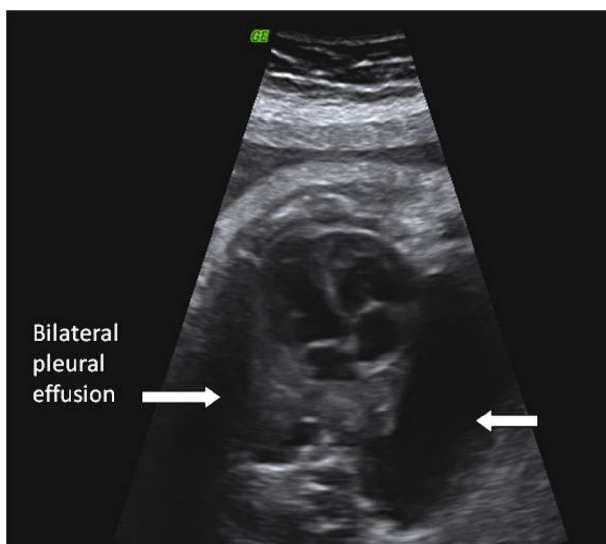


Fig. 1. Prenatal ultrasound of Case 1 at 34 weeks of gestation shows bilateral pleural effusions.

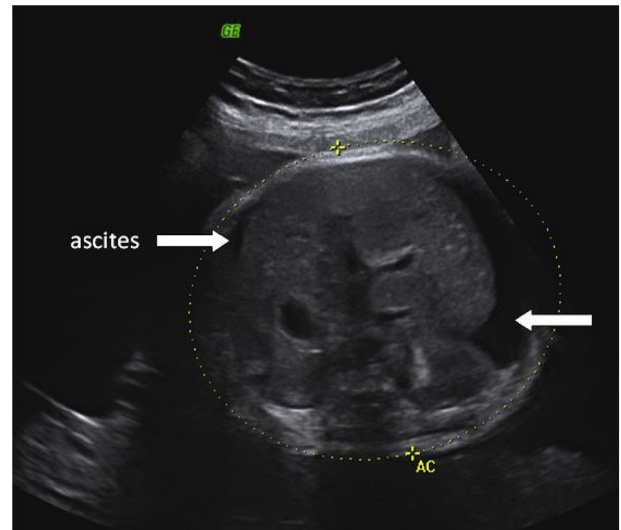


Fig. 2. Prenatal ultrasound of Case 1 at 34 weeks of gestation shows fetal ascites.

invasive screening programs and diagnostic procedures, some pregnant women choose to forgo prenatal diagnosis of Down syndrome in early pregnancy. Our cases provide evidence that fetal pleural effusion is an important sonographic marker in late gestation and especially in pregnancies without prior fetal Down syndrome screening and invasive diagnostic procedures in early gestation.

The incidence of chromosomal abnormalities associated with fetal pleural effusion has been reported to range from 6.8% to 81.8% (Table 1) [3,4,8–13]. Table 1 shows a higher incidence of chromosomal abnormalities in the first trimester than in the second and third trimester. For example, Hashimoto et al. [4] reported an incidence of 81.8% (9/11) chromosomal abnormalities associated with first-trimester pleural effusion. Conversely, fetal pleural effusion occurring in the second and third trimesters has a lower incidence of chromosomal abnormalities. For example, Blott et al. [9] reported an incidence of 9% (1/11) and Rodeck et al. [10] reported an incidence of 12.5% (1/8) in second-to-third-trimester fetal pleural effusion. According to Table 1, the overall incidence of chromosomal abnormalities associated with fetal pleural effusion is estimated to be 30.9% (148/478). This estimation is similar to the incidence reported by Nicolaidis et al. [8], Waller et al. [3] and Ruano et al. [12]. Table 1 also shows that the most common chromosomal abnormality associated with fetal pleural effusion is Turner syndrome which is followed by Down syndrome and Edwards syndrome. The incidence of fetal pleural effusion associated with chromosomal abnormalities has previously been reported in several review studies without complete cytogenetic data. In a review of 124 cases from 38 reports with fetal pleural effusion, Weber et al. [14] reported that at least four cases (3.2%) had Down syndrome. In a review of 82 cases from 31 reports with fetal pleural effusion, Hagay et al. [15] reported that at least four cases (4.9%) had Down syndrome. In a review of 147 cases from three reports and an additional six cases in their study, Achiron et al. [2] reported that at least nine cases (9/153 = 5.8%) had chromosomal abnormalities including one case with Turner syndrome and eight cases with Down syndrome.

The frequency of fetal pleural effusion in fetuses with Down syndrome is approximately 1% [16]. The proposed mechanisms of pleural effusion associated with Down syndrome include lymphatic dysplasia, transient abnormal myelopoiesis (TAM), and alveolar hypoplasia [17–21]. Moerman et al. [17] presented seven cases with bilateral chylothorax of which the perinatal autopsies of all the

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