**Objective**
We aimed to report a case of premature constriction of the fetal ductus arteriosus associated with congenital cytomegalovirus (CMV) infection.

**Methods**
Description of a case of premature constriction of the fetal ductus arteriosus associated with congenital CMV infection. A brief literature review is provided.

**Results**
A 27-year-old pregnant woman (G1P0) was referred to our service due to the presence of multiple anomalies on the first trimester ultrasound and suspicion of Edwards syndrome (trisomy 18). The first trimester screening had shown a nuchal translucency of 0.6 mm, nuchal edema, single umbilical artery, lateral ventricles measuring 1 cm, pericardial effusion, and ascites. The exam performed in our service confirmed those findings and allowed the additional identification of cardiomegaly with enlarged right chambers and left axis deviation, and premature constriction of the fetal ductus arteriosus, with mild hemodynamic repercussion. IgG testings for rubella, CMV and herpes simplex 1 (HSV-1) were positive. She had no history of usage of any medication during pregnancy. Cesarean section was performed, and the newborn presented Apgar scores of 8 in the first minute and 9 in the fifth minute, with a birth weight of 3,110 g, no dysmorphism, and normal abdominal ultrasound. Brain ultrasound, however, showed small calcifications in the thalamus and basal ganglia, and a slight enlargement of the lateral ventricles. Post-natal blood testing for CMV were IgG-reacting only, but urine PCR was positive. This finding led to the diagnosis of congenital CMV infection.

**Conclusion**
Premature closure of the fetal ductus arteriosus is a rare condition that has been previously described as idiopathic or secondary to structural lesions and use of medications. CMV infection is considered the most common congenital infection, occurring in 0.1 to 2% of all live births. Congenital CMV infection is more commonly associated with persistence of the ductus arteriosus, and not its premature closure, as observed in our case. We did not find any other report in the literature presenting such association. Our case highlights the importance of urinary CMV PCR as a screening tool for cases with clinical suspicion and non-confirmatory blood serology.