Case reports involving the first two cases of Congenital Zika Syndrome confirmed by RT-PCR. The cases were followed from the gestation through serial ultrasound (GE E8). Amniocentesis was performed at week 28 and Magnetic Resonance at week 29. Postpartum outcomes were also monitored. **Case 1** – A physical therapist, 33 years old, exanthema with seven weeks of pregnancy. Neuroimaging findings: microcephaly, lissencephaly, subcortical calcifications, cerebellar vermis and corpus callosum hypoplasia, increased interhemispheric space, and reduced brain volume. Zika virus was detected by RT-PCR in amniotic fluid. Newborn female, cephalic perimeter of 30.5 cm, Apgar at 1st and 5th of 9 and 10. The child received therapy at the Professor Joaquim Amorim Neto Research Institute until the present. The Gross Motor Function Measure was applied with 16 (median of 116), 23 (median of 170) and 34 (median of 206) weeks of life, level II by GMFCS, without seizures, walking and attending to school.

**Case 2** – A 23 years old presented rash with 18 weeks. Neuroimaging findings: severe and asymmetric ventriculomegaly, lissencephaly, absence of thalamus, severe cerebellar and cerebellar vermis hypoplasia, unilateral microphthalmia, cataract and arthrogryposis. Newborn male, cephalic perimeter of 36.5 cm and Apgar 3/3. He died at 10 hours of life. The presence of Zika virus had already been detected in the amniotic fluid by RT-PCR and the presence of viral material was confirmed after necropsy in brain tissue, meninges and kidney.

**Histopathological study evidenced an important reduction of the cerebral parenchyma, important hypoplasia and calcifications in Brain stem, with presence of lymphocytes and histiocytes in deep gray nuclei, histiocytes and brainstem.**