Since the decline in new cases of infection by insect/vector, congenital Chagas disease has become more relevant in the transmission of Chagas disease. Treatment with benznidazole significantly reduces the parasitemia, which constitutes an important factor linked to vertical transmission. The objective of this study was to evaluate whether treatment with benznidazole previously administered to women of childbearing age can prevent or reduce the incidence of new cases of congenital Chagas disease. An historical cohort study that included all women in reproductive age (15-45 years) assisted in our center was designed. We included 67 mothers with chronic Chagas disease; 35 women had not been treated prior to pregnancy, 15 had been treated prior to pregnancy and 17 gave birth prior and after treatment with benznidazole. Eight mothers gave birth to 16 children with congenital Chagas disease (8/67, 12%). The prevalence of congenital Chagas was 16/114 (14%) children born to untreated mothers and 0/42 (0%) children born to benznidazole- treated mothers, p=0.01. No significant differences were observed in clinical, serologic, epidemiological or socioeconomic baseline variables between mothers with and without children born with congenital Chagas. A 32% conversion rate to negative serology was observed in benznidazole-treated women after long-term follow up. Antiparasitic treatment administered to women in reproductive age can prevent the occurrence of congenital Chagas disease.