Endometriosis is one of the most common benign gynecological diseases affecting women during reproductive age; it is characterized by the presence and growth of ectopic endometrial tissue outside the endometrial cavity. This complex disease is frequently associated with infertility and pelvic pain. Given the relationship and the apparent importance of the role that neurotrophins play in the reproductive system, and in particular Brain-derived Neurotrophic Factor (BDNF) which is involved in both the central and peripheral pain pathways, we were interested in determining whether the presence of endometriosis is associated and related with plasma and follicular fluid variation of BDNF. We determined BDNF level in plasma and in follicular fluid from infertile women with endometriosis and fertile women without the disease. BDNF plasma levels were significantly higher in patients affected by endometriosis than in control women (p<0.001). After surgery this level decreased significantly (p<0.001), ranging within the values of control women in follicular phase. In follicular fluid, BDNF values were significantly lower in infertile women caused by endometriosis than in infertile women for male factors (p<0.001). These data suggest that neuroinflammatory reactions in endometriosis could have a neuroprotective effect and support the hypothesis that BDNF represents an important link in the networks of human homeostasis, thus providing an early marker for patients affected by endometriosis. Moreover, low BDNF levels in follicular fluid may reflect an altered ovary production and may be a marker of poor oocyte quality and poor fertility in women suffering from endometriosis.