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Very long-term outcomes of pediatric patients treated for optic pathway gliomas: A longitudinal cohort study

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Abstract

Background: Optic pathway gliomas (OPG) represent 5% of childhood brain tumors. Successive relapses lead to multiple treatments exposing to late complications.

Methods: We included patients treated at Gustave Roussy (GR) between 01.1980 and 12.2015 for OPG, before 18 years-old and alive at 5 years from diagnosis. Mortality and physical health conditions data were extracted from medical data files and updated thanks to the GR long-term follow-up program and French national mortality registry for patients included in the French Childhood Cancer Survivor Study.

Results: We included 182 5y-OPG-childhood survivors in the analysis (sex-ratio M/F 0.8, 35% with NF1). With a median follow-up of 17.2y (range=5-41), we registered 82 relapses, 9 second malignancies and 15 deaths as first events after 5 years, resulting in 20-y conditional overall survival (C-OS) and late events-free survival (LEFS) of 79.9% (95%CI=71-86) and 43.5% (95%CI=36-51) respectively. NF1 (Hazard ratio HR=3, 95%CI=1.4-6.8), hypothalamic involvement (HR=3.2, 95%CI=1.4-7.3), and radiotherapy (HR=2.8, 95%CI=1.1-6.7) were significantly associated with C-OS in multivariable analyses. Ninety-five percent of 5y-OPG survivors suffered from any health condition, especially visual acuity "<1/10" (n=109), pituitary deficiency (n=106) and neurocognitive impairment (n=89). NF1 (HR 2.1) was associated with precocious puberty. With a median time post diagnosis of 4.2 years, 33 cerebrovascular events were observed in 21 patients.

Conclusion: Late relapses, second malignancies and cerebrovascular diseases are severe late events resulting in premature mortality. Morbidity is high and needing after-cancer care to improve quality of life. Risk factors could be considered to better stratify long-term follow-up.

Keywords: long-term outcome; optic pathway glioma; pediatric.

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