Fibroblast growth factor receptor (FGFR) alterations are prevalent in human cancers. In a large Canadian population-based cohort, encompassing over 1,300 pediatric and Adolescent and young adult (AYA) gliomas, FGFR mutations occurred in 10% of pediatric low-grade gliomas (LGG) and 20% of IDH-WT AYA LGGs. Here we assembled a large multi-institutional cohort including more than 300 mutated gliomas, aged 6 months to 67 years, 53% of which were LGG. High-grade gliomas were more common with increasing age; over 90% of pediatric cases were LGGs, compared to 30% in the AYA group. Fusions, predominantly seen in the pediatric group (70% of LGGs), contrast with FGFR3-TACC3 fusions frequent found in adult high-grade gliomas. Interestingly, FGFR mutations sometimes existed as sole drivers or co-occurred with other alterations (~30%), it is unclear how this alters tumor behavior, although gain of additional alterations could be implied in malignant transformation and poor prognosis. Despite higher incidence of high-grade gliomas in AYAs, their progression-free survival was significantly better than that of pediatric LGGs (p=0.007), largely due to the prevalence of hemispheric tumors amenable to gross total resection. Approximately 30% of our LGG cohort underwent adjuvant therapy; with available data for 28 patients, objective responses were noted in 2/12 to chemotherapy, 2/9 to MEK inhibitors, and 3/7 to FGFR inhibitors, respectively. With the increasing promise of precision targeted therapy, molecular profiling and assessment for FGFR alterations is essential. Although encouraging, further studies are needed to assess the benefit of FGFR inhibition in these patients.