CASEREPORT: Neurofibromatosis type 1 (NF1) associated pilocytic astrocytomas occur most frequently in the optic pathway, hypothalamus and brainstem. Astrocytomas of the corpus callosum are relatively rare in this population (about 15% of NF1 associated intracranial tumors) and have not been well characterized. We report a 12 year old boy with a recurrent NF1 associated pilocytic astrocytoma of the corpus callosum. Our patient has ADHD, Tourette’s syndrome and hyperplasia of the left optic nerve and orbit. He has no history of seizures or any focal neurological deficits. MRI done to investigate new onset headaches showed interval development of a 1.8 X1.2 cm T2 hyperintense mass in the central corpus callosum. There was no surrounding vasogenic edema. No other enhancing lesions were seen in the corpus callosum. Pathological examination after sub-total resection showed the tumor to be well circumscribed with cells having long cytoplasmic processes and eosinophilic granular bodies consistent with a WHO grade 1 pilocytic astrocytoma but with infiltrative edges suggestive of WHO grade 2 diffuse astrocytoma. The tumor was GFAP(+) , Neu-N(-) and Ki-67= 1%-7%. A few cells were p53(+) , CD45(+) and CD68(+). Post-operative MRI showed enhancement along the surgical bed and follow up MRI done 8 months later showed progression of residual tumor. At repeat surgery all grossly apparent tumor was resected. Pathological examination showed moderate cellularity, nuclear pleomorphism, no mitotic activity, no necrosis, GFAP (+ ), S-100(+), Neu-N(-) and Synaptophysin(-). Ki-67 positivity was minimal. MRI 3 months later showed a 7 mm enhanced focus in the left paramedian posterior body of the corpus callosum which has remained stable over 6 months. The child is being closely monitored. The tumor grade/proliferative potential of corpus callosal pilocytic astrocytomas in NF1 patients may be different from astrocytomas in other locations and merit close monitoring and further study.