BACKGROUND: Medulloblastoma is the most common malignant brain tumor in pediatric population. Standard treatment combines surgery, risk-adapted craniospinal irradiation and chemotherapy. Given the well-known cognitive sequelae in young children associated to radiation, strategies in infants try to avoid upfront radiation. A significant number of patients treated during infancy will ultimately relapse. Re-irradiation in relapse setting has proven to be effective in disease control (survival depending on several factors) although there is a concern in exceeding radiation tolerance and risk of toxicity. METHODS: Herein, we describe the case of male patient that presented at 1 year of age with a non-metastatic anaplastic medulloblastoma that was completely resected. He received 3 cycles of adjuvant chemotherapy (as per Head Start II), local relapse was detected in tumor bed during evaluation and was completely resected, afterwards he received myeloablative chemotherapy with stem cell rescue and posterior fossa radiation therapy (54 Gy). At 8 months off therapy patient developed spinal relapse in imaging with negative cytology, then treated with spinal irradiation 36Gy without adjuvant chemotherapy. He remained disease free during 12 months, then tumor recur at the right cerebellopontine angle, surgery was complete and there was no evidence of distant disease. Patient received adjuvant local radiation with 30.6Gy followed by chemotherapy (bi weekly bevacizumab + irinotecan). Cumulative maximal dose to brainstem was 84Gy. 2 months after radiation patient presented with extreme sleepiness and imaging findings compatible with brainstem radiation necrosis, symptoms resolved promptly after scheduled bevacizumab. Patient is currently 6-year-old and remains free of disease after 3 years off therapy. CONCLUSION: Medulloblastoma treatment in infants is challenging because survival is not the only goal. Future clinical trials will help to better tailored treatment based on molecular biology. Re-irradiation in medulloblastoma has been described as well tolerated but clinician should be aware of its complications and management.