Central nervous system involvement in Acute Myeloid Leukemia is uncommon, and management and outcomes are not well defined. AML patients, in the past 5 years, with CSF positivity for malignant cells or direct CNS/spinal cord involvement at presentation were included in this study. Patients with paraspinal masses, intra cranial bleed, CNS involvement at relapse, unconfirmed CNS involvement were excluded. Eight patients fulfilled the inclusion criteria, 4 of which were paediatric patients. Cytogenetics were t(8:21), t(8:21) +8, t(8:21) +8 +19, t(9:22) in 1 patient each with rest 50% patients having normal cytogenetics. CSF was positive for malignant cells in 3/7 patients. Imaging in 6/8 patients showed leptomeningeal involvement in 2 patients, cavernous sinus and falk cerebri involvement in 1, paravertebral deposits and cord enhancement in 1, and paravertebral and infratemporal deposits in another, and was normal in 1 patient with 3rd cranial nerve palsy. All patients were treated with 3+7 induction followed by HIDAC with IT therapy and RT in 2/8 patients. 6/7 treated patients showed near complete resolution of CNS involvement after chemotherapy. 1 patient with paraplegia had persisting spinal cord lesions despite 2 induction chemotherapies, IT and RT and subsequently had bone marrow relapse. One patient underwent HSCT and relapsed within 3 months and expired. Four other patients also relapsed – one CNS relapse, 2 CNS and BM relapse, and 1 BM relapse. Currently only 2/8 patients are alive. Presentation of CNS involvement in AML is variable with a high rate of disease relapse and mortality.