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Diabetes Insipidus as the initial presenting symptom in an Adult with Langerhans Cell Histiocytosis

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There is paucity of information on Langerhans Cell Histiocytosis (LCH) in adults because of its rarity in this age group. We report a case of Central Diabetes insipidus as the initial presentation in an adult patient with LCH. To our knowledge, this is the 2nd adult case of LCH presenting with Diabetes insipidus reported in literature in the Philippines.

A 27 year-old female presented with polyuria and polydipsia, initially thought to be part of normal pregnancy, however symptoms did not improve post-partum. She deferred consult until she developed recurrent headaches associated with left hemifacial anesthesia 7 years later. Initial CT scan showed left parietal calvarial tumor, no other intracranial lesion was reported. Diabtes mellitus was excluded during this time. Craniectomy, excision of the tumor and cranioplasty was carried out. Histopathology and immunostaining with S100 and CD1a confirmed the diagnosis of Langerhans Cell Histiocytosis. Central diabetes insipidus was confirmed after water deprivation test and patient was maintained on Desmopressin with improvement in polyuria. Subsequent endocrine workup revealed isolated hypothyroxinemia. Thyroxine levels improved after initial courses of chemotherapy.

Langerhans cell histiocytosis (LCH)is a rare disorder of granulomatous deposition at multiple sites within the body1 known to involve the hypothalamo-pituitary axis (HPA). In particular, development of diabetes insipidus may precede the diagnosis of LCH by years in some reported cases. In the absence of reported lesion in the HPA in imaging, it is prudent to include workup of anterior pituitary hormone to rule out other deficiencies which could contribute to its symptomatology.