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CORRESPONDENCE

Letter to the editor: The "Marabou-feather wing" sign in hypoparathyroidism

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Hypoparathyroidism is an endocrine-deficient disorder characterized by an absence or elimination of parathyroid hormone (PTH) and a derangement of the calcium homeostasis causing hypocalcemia. The condition can be inherited or acquired, with thyroidectomy comprising the second most common etiology of PTH deficiency in adults [1]. In PTH-deficient hypoparathyroidism, hypocalcemia coupled with hyperphosphatemia is responsible (among other manifestations) for the ectopic deposition of insoluble calcium phosphate complexes in the brain. Awareness of the neurological manifestations of hypoparathyroidism affecting the basal ganglia is important because abnormalities can be associated with serious morbidity and significant socioeconomic costs [1]. Awareness of hypoparathyroidism also is important because such awareness facilitates differentiation from other entities that can produce similar radiologic findings but have very different treatments and prognoses, such as Fahr's disease, pseudohypoparathyroidism, primary hyperparathyroidism, hyperthyroidism, pure Parkinsonism, and dementia. We report our observations in secondary hypoparathyroidism featuring MR imaging changes in the basal ganglia that resemble the appearances of the Marabou bird fine feathered wings, for which we have coined the "Marabou-feather wing" sign. This association suggests this sign can be a clue to the diagnosis of PTH-deficient hypoparathyroidism associated with dysregulation of calcium homeostasis and cerebral changes.

A 54-year-old woman with Parkinsonian symptoms presented with a sudden episode of impaired sensorimotor function of the right upper arm and dysarthria that had resolved spontaneously earlier that day. She had total thyroidectomy 10 years earlier, for which she received thyroid hormone replacement therapy. The patient developed extrapyramidal symptoms a few years later that had been slowly deteriorating. She had tremor at rest. Laboratory tests showed hypocalcaemia (1.28 mmol/L) and undetectable level of parathyroid hormone (PTH) < 0.4 pg/ml. There was vitamin D deficiency (17ng/ml) and elevated serum phosphorus (1.8 mmol/lt). While in the ER, the patient became confused and developed generalized epileptic seizures. CT images of the brain demonstrated widespread dense calcifications, mostly in the basal ganglia (Fig. 1A). To exclude transient ischaemic attack (TIA) causing her sudden weakness and slurred speech, she underwent cerebral MRI. Diffusion weighted sequences eliminated diagnosis of acute ischemic stroke. MRI revealed areas of hyperintense signal on all sequences, in the head of caudate nucleus, and the internal capsule and the thalami (Fig. 1B). The patient was diagnosed with chronic (post-thyroidectomy) hypoparathyroidism and cerebral calcifications, most prominent in the basal ganglia, accounting for disturbances in motor control and cognitive function. Symptomatic hypocalcemia was rapidly corrected, and hypoparathyroidism was treated with oral calcium supplementation and daily vitamin D. Twelve weeks after the initiation of therapy her calcium, phosphorus, and vitamin D levels had been restored to the lower end of the normal range and the extrapyramidal symptoms had improved.

Total thyroidectomy is one of the most common surgical procedures that may also be complicated by transient or permanent hypocalcemia due to direct injury to the parathyroid glands, or inadvertent devascularization of these delicate glands. Of note, reported data indicate post-operative parathyroid failure rates that range from 19% to 38% [1].

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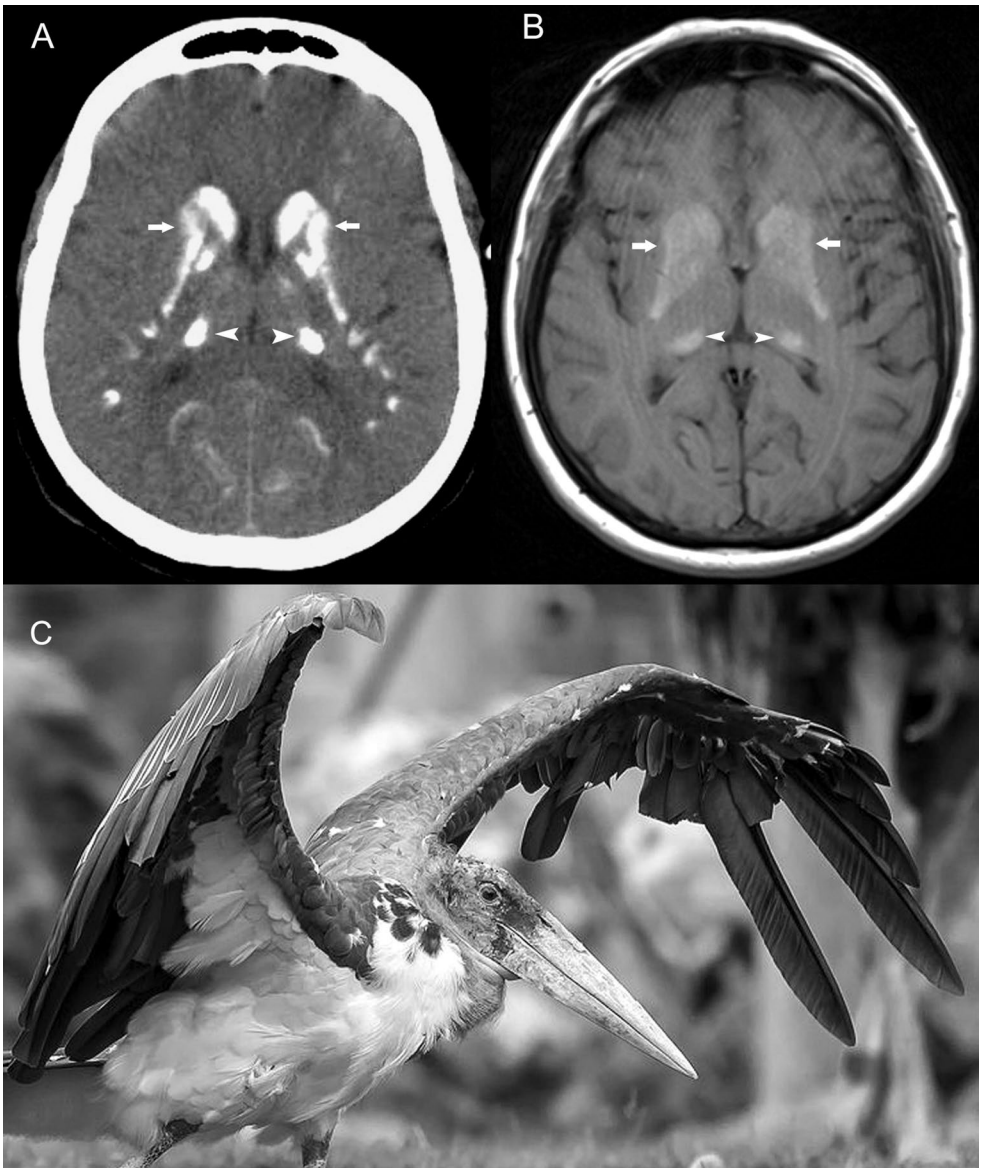

Fig. 1 Images in a 54-year-old woman with total thyroidectomy on admission due to dysarthria and weakness. (**A**) Axial CT scan of brain shows massive calcifications in the bilateral basal ganglia (arrows) and the thalami (arrowheads). (**B**) Corresponding T1-weighted MR image demonstrates bilateral, symmetrical abnormal high signal intensity in basal ganglia with a fine, feathery wing appearance representing the "Marabou-feather wing" sign. (Sections are at approximately the same level). (**C**) The Marabou stork. Delicate, airy feathers are used for years to embellish luxurious garments. (Image courtesy of Hoan Luong. Reproduced with permission from www.pinterest.com/pin/66498531997036753)

The prototypical brain lesion associated with chronic hypoparathyroidism however, is calcification of the basal ganglia. Commonly CT images exhibit bilateral, symmetric cerebral calcifications with corresponding hypointense signal on MR images [2]. We documented hyperintense areas in the head of the caudate nucleus and the internal capsule on T1-weighted MR images presumably representing ectopic deposition of proteins and/or mucopolysaccharides in the brain [3]. We postulate that these deposits in turn, bind the mineral ions (calcium or other metallic ions) forming colloid mixtures and brain calcifications. Previous researchers have found that the caudate nuclei are able to express various osteogenic molecules—mostly proteins, similar to those involved in the mineralization of bone [4]. In addition, lamellar deposition of calcium-hydroxyapatite crystals has been reported in the basal ganglia of patients with hypoparathyroidism suggesting an active osteogenic process [4, 5]. Although the etiology and pathogenesis of preferential involvement of the basal ganglia is unclear, the abnormal, symmetrical nebulous accumulation of colloids in metabolically active central grey matter forming a pair of wings, is reminiscent of the delicate fuzzy feathers of the Marabou African stork (taxonomy, *Leptoptilos crumeniferus)* wings (Fig. 1C).

The Marabou-feather wing sign described herein is non-specific for PTH-deficient hypoparathyroidism manifesting in the brain. Besides, intracranial calcifications may relate to numerous medical conditions, and aging [4]. Although we have no histologic confirmation regarding the composition of deposits in the basal ganglia, our observations with MR imaging of colloid deposition are inferred from previous advanced immuno- histochemistry/genetic studies

identifying molecules related to active osteogenesis (rather than metastatic or dystrophic deposition of minerals) that may predispose the basal ganglia for calcification [4, 5]. Although non-pathognomonic, the Marabou-feather wing sign in the basal ganglia of patients with hypoparathyroidism should be considered a benign entity and not mistaken for a more ominous hyperintense brain lesion such as cerebral hemorrhage, a fat-containing tumor or some multiple sclerosis plaques.

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Data availability No datasets were generated or analysed during the current study.

Declarations

Competing interests The authors declare no competing interests.

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