Bimanual synkinesia in Kallmann's syndrome

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DESCRIPTION

A man aged 20 years presented with complaints of small penile size and absence of both testes in his scrotum. The same was documented at birth but no intervention was done ever since. He was tallest in the family and had two normal siblings. He had poor development of secondary sexual characters. He also had history of anosmia since childhood. His arm span was more than his height by 8 cm and the upper:lower segment ratio of his body was 0.76 (normal >0.9) suggesting a eunuchoid habitus. He had micropenis and both the testes were not palpable in the scrotum or inguinal canal (figure 1). On further examination, he had asymmetric bimanual synkinesia (video 1). He had no other neurological features.

His baseline hormonal evaluation revealed a total testosterone of 19.5 ng/dL with undetectable luteinising hormone (LH) levels, while other anterior pituitary hormones were normal (including free T4, cortisol, prolactin). His peak LH was 0.49 mIU/mL after a 500 µg leuprolide stimulation test. Ultrasonography (USG) showed both the testes in the inguinal canal, right measuring 1.4/0.5 cm and left was 1.4/0.7 cm. No renal abnormality was found in USG. His audiometry was normal. Coronal T2-weighted MRI of the brain showed hypoplastic right olfactory sulcus and olfactory bulb. The left olfactory sulcus

Video 1 Video showing asymmetrical bimanual synkinesia, initially on the right followed by left.

Figure 2 High-resolution coronal T2-weighted MRI of the patient showing hypoplastic olfactory bulbs in both the side (horizontal arrows). The right olfactory sulcus is hypoplastic (coloured vertical arrow). Olfactory sulcus is absent on left side (white vertical arrow).

was absent (figure 2). He was diagnosed with Kallmann's syndrome.

Bimanual synkinesia, also known as mirror movement, is a classic neurological finding in patients with Kallmann's syndrome, particularly the X linked variety, which is associated with KAL-1 mutation.¹ However, it can also be found in

Figure 1 Image of the patient showing micropenis and poorly developed scrotum.

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Learning points

- ▶ Kallmann's syndrome is characterised by the association of congenital hypogonadotropic hypogonadism and anosmia or hyposmia.
- ▶ Bimanual synkinesia is a very important clinical clue to identify Kallmann's syndrome as the aetiology of hypogonadism.
- ▶ Absent or hypoplastic olfactory sulcus and bulbs is a key radiological pointer towards Kallmann's syndrome.

acquired disorders like stroke, parkinsonism or dystonic disorders. It has been hypothesised that abnormal neurological circuit or functional alteration in planning and execution is responsible for this mirror movement.² Although mirror movement may not be present always, it should be looked for in hypogonadal men. If present, mirror movement can serve as an important clinical clue for diagnosis of Kallmann's syndrome.

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