Persistent singultus as presenting symptom of syringobulbia

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DESCRIPTION

A 22-year-old man presented with persistent hiccups (singultus) since 15 days with giddiness, imbalance while walking and heaviness in the right upper limb since 10 days. On clinical examination the patient had left ataxia with reduced sensation to pinprick over right C5–C7 segments. The patient was suspected to have a neurogenic cause of singultus hence was referred for MRI of the brain and spine.

MRI demonstrated syringobulbia as a slit-like hyperintensity on T2-weighted image (figure 1A, B) in the right dorsal aspect of the medulla which suppressed on fluid attenuated inversion recovery (figure 1C, D). MRI of the cervical and dorsal spine (figure 2) demonstrated syringomyelia from the C2–D2 level. Peg-like cerebellar tonsils displaced into the upper cervical canal through the foramen magnum (figure 2) suggestive of a Chiari I malformation.

Acute singultus (hiccups) is usually a benign transient phenomenon usually caused by overdistension of the stomach, chemical stimulation (such as alcohol) or sudden changes of gastrointestinal temperatures. It involves repeated myoclonic, synchronous, involuntary contractions of the diaphragm and inspiratory intercostal muscles associated with an upper airway closure causing a peculiar sound.¹

Hiccups are classified according to their duration: acute hiccups that last up to 48 h, persistent hiccups that last for over 48 h and intractable hiccups are defined as hiccups that last more than 1 month.²

Persistent and intractable singultus may cause severe discomfort, decreased physical strength and depression if left untreated. Persistent and intractable singultus may indicate an organic disorder and should be investigated based on history and physical examination with imaging.

Figure 1 MRI of the brain: axial T2-weighted image (A and B) showing slit-like hyperintensity (arrows) in the right dorsal region of the medulla which is suppressed on fluid attenuated inversion recovery (C and D).

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Figure 2 Sagittal short tau inversion recovery images of cervicodorsal spine demonstrating tonsillar herniation (white arrow) into the upper cervical canal, syringobulbia seen as hyperintensity in the medulla (dashed arrow) and syringomyelia of the cord from extending from C2-D2 level.

Neurogenic causes of persistent or intractable singultus include structural or functional disorders of the medulla (involving the region of vagal nuclei and nucleus tractus solitarius) and of the afferent or efferent nerves to the respiratory muscles.³ Structural lesions of the medulla such as infarction in the territory of the posterior inferior cerebellar artery, tumour, tuberculoma, abscess, syrinx, haematoma and demyelination can cause persistent singultus.³ MRI plays an important role in the evaluation of persistent singultus and diagnosis of brainstem lesions of which syringobulbia due to the Chiari I malformation is a treatable cause.

In our case the cause of persistent singultus can be attributed to the syringobulbia due to Chiari I malformation. The patient underwent foramen magnum decompression and his symptoms were alleviated postsurgery.

Learning points

- In persistent or intractable singultus (hiccups), a brainstem lesion should be suspected, although the incidence is low.
- MRI plays an important role in the evaluation of persistent singultus and diagnosis of brainstem lesions.
- Syringobulbia due to Chiari I malformation represents a surgically treatable cause of intractable or persistent singultus.

Competing interests None.

Patient consent Obtained.

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