Use of Osteopathic Manipulative Treatment to Manage Recurrent Bouts of Singultus

Benjamin Seidel, DO
Gina Benaquista Desipio, DO

From the Department of Physical Medicine and Rehabilitation at Moss Rehabilitation Hospital in Elkins Park, Pennsylvania (Dr Seidel), and the Department of Physical Medicine and Rehabilitation at Thomas Jefferson University Hospital in Philadelphia, Pennsylvania (Drs Seidel and Benaquista Desipio).

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Address correspondence to Benjamin Seidel, DO, Thomas Jefferson University Hospital - Physical Medicine and Rehabilitation, 111 S 11th St, Philadelphia, PA 19107-4824.

E-mail: bseidel44@gmail.com

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Singultus, or hiccups, are involuntary spasms of the diaphragm that in most cases are harmless and self-limited. Treatments are reserved for those cases that persist, and current options include pharmacotherapeutics, complementary methods (such as acupuncture), and osteopathic manipulative treatment. A 32-year-old woman with stiff person syndrome and concurrent aminoacidopathy in the setting of acute inpatient rehabilitation was experiencing daily bouts of singultus, ranging from 20-minute to 5-hour durations. Osteopathic manipulative treatment at the onset of spasm resulted in immediate cessation of and further suppression of singultus for approximately 12 to 24 hours. Overall, there was a noted reduction in singultus frequency, duration, and intensity, as well as better tolerance of physical and occupational therapy. The authors theorize that OMT could be a useful adjunct to, or replacement of, pharmacologic interventions for singultus, especially when pharmacologic therapies have failed.

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Singultus, or hiccups, is defined as "an involuntary, intermittent, spasmodic contraction of the diaphragm and intercostal muscles."[1] The current definitions divide singultus into 4 categories: acute, persistent, chronic, and intractable. Acute singultus is defined as hiccups lasting fewer than 48 hours; persistent, more than 48 hours; chronic, more than 7 days; and intractable, more than 1 month.[2,3] We describe a patient with recurrent bouts of presumed centrally caused singultus that responded to osteopathic manipulative treatment (OMT).

Report of Case

A 32-year-old woman was admitted to an acute rehabilitation hospital at a large academic medical center after receiving a diagnosis of ileus and experiencing prolonged nausea and vomiting. The patient had a history of stiff person syndrome (glutamic acid decarboxylase [GAD]-65 antibody negative), an aminoacidopathy of mitochondrial origin diagnosed in 2008, and associated postural tachycardic syndrome and full body spasms. Before the current hospitalization, the patient had no history of gastrointestinal symptoms nor singultus.

The onset of singultus occurred early in her hospitalization, manifesting as recurrent bouts most frequently when the patient was in an upright position or when she forward-flexed either humerus. These episodes would last 20 minutes to 5 hours before spontaneous cessation. Various medications, including baclofen, ondansetron, metaclopramide, and carbemazepine, were unsuccessful in reducing the duration, frequency, or intensity of singultus. Three weeks after its onset, an episode of intractable singultus developed while the patient was on a tilt table during physical therapy. An osteopathic physician (B.S.) was consulted.

The physician conducted an osteopathic structural examination and found tissue texture changes, asymmetry, and restriction of motion or tenderness in the neck, T1-T4, T10-T12, and sacrum and pelvic regions (*Table 1*). He then performed OMT using rib raising, diaphragmatic balanced ligamentous tension, and diaphragmatic doming of the left hemidiaphragm,[5] with instantaneous cessation of singultus. Singultus recurred the following day after a similar position change during therapy, and OMT was performed again with similar results. Four days later, during a period of singultus remission, we performed another osteopathic structural examination (*Table 2*), which showed persistence of the cervical, upper thoracic, thoracolumbar, and sacral dysfunction, with the addition of cranial and abdominal findings. Myofascial release and osteopathic cranial manipulative medicine were applied to the occiput, neck,

thoracic spine, sacrum and pelvis, and superior mesenteric ganglion. Tissue texture, asymmetry, restriction of motion, and tenderness (ie, the TART criteria) resolved, and the cranial rhythmic impulse had improved. After OMT, therapists noted that the patient had better tolerance of upright posture and forward flexion of either humerus. She was able to achieve a modified level of independence, with self-feeding, upper extremity dressing, and independent sitting, which she was previously unable to tolerate. The patient was not followed up as an outpatient.

Discussion and Review of the Literature

In most cases, singultus is observed to occur as unilateral spasms of the left hemidiaphragm.[6] To our knowledge, little is known regarding the underlying pathophysiology.[7] In one case study,[8] the physiologic reflex arc was reported to involve an afferent limb of the phrenic and vagus nerves, central mediators, and a subsequent efferent limb involving the phrenic nerve and accessory connections to the glottis and intercostal muscles. The central connections have not been well delineated but may involve the medulla oblongata, reticular formation, phrenic nerve nuclei, and hypothalamus.[8] As such, central nervous system disorders would be associated with a small number of singultus cases.[9] In the current case, both forward flexion of either humerus and sitting upright exacerbated the singultus. Although speculative, it may have been secondary to engagement of the serratus anterior muscle during forward flexion and the paraspinal muscles when sitting upright, all of which share origins on the rib cage.

The suggested treatment for patients with singultus depends on the cause; therefore, a reasonable attempt should be made to eliminate the cause. For example, if a particular drug (eg, dexamethasone) is thought to be the cause, treatment would involve cessation of the medication. If no cause can be determined, various medications used to stimulate or suppress the central nervous system have demonstrated efficacy. Sources in the literature have supported the use of chlorpromazine as the drug of choice for empiric therapy.[10,11] Chlorpromazine, a phenothiazine antipsychotic medication, is generally well tolerated at the low doses used to treat singultus. Other medications include metaclopramide, baclofen, anticonvulsants, tricyclic antidepressants, methylphenidate, quinidine, olanzapine, and amantadine.[10] In addition, several case and observational studies have shown positive outcomes with acupuncture.[12,13] One Chinese study compared acupuncture with methylphenidate in 80 patients with singultus after cerebrovascular accident, finding superior results with acupuncture and traditional cupping techniques.[10] A 2013 systematic review[14] failed to demonstrate conclusive efficacy of pharmacologic or nonpharmocologic interventions and noted the following:

> [T]here is insufficient evidence to guide the treatment of persistent or intractable hiccups with either pharmacological or non-pharmacological interventions. The paucity of high quality studies indicate a need for randomised placebo-controlled trials of both pharmacological and non-pharmacological treatments.

To our knowledge, there have been no studies investigating the use of OMT in patients with recurrent neurogenic singultus, although its use as treatment for patients with singultus has been reported.[15] Physical maneuvers to alleviate singultus have a long history worldwide[7] and vary widely in technique. The research is peppered with insufficient evidence and case reports. In the mid-1800s, Sir William Osler proposed applying traction to the tongue, reporting a benefit in patients with chronic singultus.[16] Most maneuvers aim to decrease the direct irritation to the diaphragm (eg, pulling knees to chest), increase vagal tone to the diaphragm (eg, Valsalva maneuver), interrupt respiration with breath holding, or stimulate the uvula or nasopharynx (eg, drink cold water). None of these maneuvers has been directly compared,

Table 1.
Osteopathic Structural Examination Form[a] and Findings on Initial Evaluation of a Patient With Recurrent Singultus

	Somatic Dysfunction								
	Presence				Severity[b]				
Region Evaluated	T	A	R	T	0	1	2	3	Findings
Head and face									No dysfunction
Neck	X	X	X				X		OA FS_LR_R, midcervical group FS_RR_R
T1-T4	X	X	X					X	Right first rib elevation, T1-T4 FS_LR_L with paraspinal spasm
T5-T9									No dysfunction
T10-T12	X	X	X	X			X		Thoracolumbar junction S_RR_R
Lumbar									No dysfunction
Sacrum/pelvis	X	X	X				X		Left on left sacral torsion
Pelvis/innominates									No dysfunction
Abdomen/other									No dysfunction
Right upper extremity									No dysfunction
Left upper extremity									No dysfunction
Right lower extremity									No dysfunction
Left lower extremity									No dysfunction

[a] Adapted from the Osteopathic Single Organ System Musculoskeletal Exam Form, from Sleszynski et al.[4]
[b] Severity scale: 0, no significant difference or background levels; 1, more than background levels, minor tissue texture, asymmetry, restriction of motion, and tenderness (TART); 2, obvious TART (especially restriction or tenderness), +/- symptoms; 3, key lesions present, symptomatic, restriction and tenderness stand out.[4]

Abbreviations: F, flexed; OA, occipitoatlantis; R_L, rotated left; R_R, rotated right; S_L, sidebent left; S_R, sidebent right.

and physical maneuvers only have grade 2C evidence (weak recommendation, low quality evidence).[1,17,18]

There have been no studies specifically looking at OMM in recurrent neurogenic singultus management, although OMM as a treatment for singultus has been reported in the past.[15] The basis for OMM in treatment of singultus stems from the tenets of osteopathic medicine. Structural dysfunctions can clearly be correlated with physiologic consequences in this case. Specifically, the tenets in this case can be applied to the hypothesized pathophysiologic basis of singultus formation. Osteopathic manipulative treatments such as diaphragmatic doming techniques stretch the fibers of the diaphragm and function similarly to breath-holding. Holding a muscle in a stretch directly prevents muscular contraction, therefore interrupting the spasm. Rib raising techniques balance the sympathetic innervation to the diaphragm along the thoracic spine, as well as reduce irritation to the diaphragm by inhibiting the inspiratory intercostal muscles. Phrenic nerve irritation from cervical dysfunction could have implications in singultus development. Treatment in such a case would involve

Table 2.
Osteopathic Structural Examination Form[a] and Findings of a Patient With Recurrent Singultus When Reassessed for Prevention of Singultus

	Somatic Dysfunction								
	Presence				Severity[b]				
Region Evaluated	T	A	R	T	0	1	2	3	Findings
Head and Face	X	X	X	X			X		Right occiput ESBR, right parietal flexion, with overall decreased amplitude of the cranial rhythmic impulse
Neck	X	X	X				X		OA FS_LR_R, midcervical group FS_RR_R
T1-T4	X	X	X					X	Right first rib elevation, T1-T4 paraspinal spasm
T5-T9									No dysfunction
T10-T12	X	X	X	X			X		Thoracolumbar junction S_RR_R
Lumbar									NA
Sacrum/pelvis	X	X	X				X		Left on left sacral torsion
Pelvis/innominates									No dysfunction
Abdomen/other	X	X	X				X		Superior mesenteric ganglionic dysfunction
Right upper extremity									No dysfunction
Left upper extremity									No dysfunction
Right lower extremity									No dysfunction
Left lower extremity									No dysfunction

[a] Adapted from the Osteopathic Single Organ System Musculoskeletal Exam Form, from Sleszynski et al.[4]
[b] Severity scale: 0, no significant difference or background levels; 1, more than background levels, minor tissue texture, asymmetry, restriction of motion, and tenderness (TART); 2, obvious TART (especially restriction or tenderness), +/- symptoms; 3, key lesions present, symptomatic, restriction and tenderness stand out.[4]

Abbreviations: ESBR, extended, sidebent, and rotated; F, flexed; OA, occipitoatlantis; R_L, rotated left; R_R, rotated right; S_L, sidebent left; S_R, sidebent right.

resolution of cervical dysfunction. By regulating the sympathetic and parasympathetic afferent and efferent limbs, increases in vagal tone may help to suppress the reflex arc. Balanced ligamentous tension techniques may reduce irritation to the diaphragm by balancing the surrounding ligaments, tendons, and muscular insertions of the diaphragm, thereby removing the diaphragm from a singultus-conducive posture. Reduction in singultus frequency, duration, and intensity might also be achieved through careful assessment of the major diaphragms in the body, with particular emphasis of the occipitoatlantal joint and cranial vault in cases of potential neurogenic origin. By balancing the major diaphragms of the body, the respiratory diaphragm would be in optimal position to participate in respiration without restriction. Specifically, manipulation of the occipitoatlantal joint would balance the vagus nerve, thus modulating innervation to the diaphragm.

Conclusion

To our knowledge, this is the first clinical observation of OMT as an effective treatment option for recurrent bouts of neurogenic singultus. Although further study is warranted, the potential implication of OMT as a treatment modality for singultus is encouraging for the patient population in whom medical therapy fails. In addition, OMT is a safe alternative to conventional pharmaceutical interventions, which may have adverse effects.[5] Future studies might compare the efficacy of OMT vs current medical therapies for singultus suppression and reduction of frequency.

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