New daily persistent headache (NDPH) is a rare, poorly understood, treatment-refractory primary headache disorder with heterogeneous symptoms. Some patients report a preceding upper respiratory tract infection, stress, or surgery. Additionally, Rozen et al\(^2\) note that hypermobility of the cervical spine may increase susceptibility to NDPH, suggesting a biomechanical component. Osteopathic manipulative treatment (OMT) has been shown to be useful for headache disorders, including migraine.\(^3,4\) The diagnostic criteria for NDPH are persistent daily headache not better classified by another headache type that has been present for at least 3 months and has a clearly remembered onset, with pain becoming unremitting within 24 hours.\(^5\) Many patients complain of generalized or unilateral head pressure, throbbing, photophobia, phonophobia, lightheadedness, vomiting, autonomic symptoms with painful exacerbations, and allodynia.\(^1\) This case report describes a patient with NDPH that resolved after OMT.

**Report of Case**

A 15-year-old girl presented to the pediatric neurology department with a 2-year history of continuous headache characterized by constant pressure in the center of her head radiating outward with a pulsatile quality, and it was intermittently sharp. She could recall the exact time of onset. The pain intensity ranged from 2 to 7 out of 10 on a Likert pain scale during the first year of the headache, but then stayed mainly at 7 in the subsequent year. Her headache log revealed that most days, her pain score was 6. The pain was associated with prominent phonophobia, facial flushing, and exhaustion, but not nausea, vomiting, or position. She could not participate in schoolwork or extracurricular activities. Exercise improved the pain but did not resolve it. Stress from schoolwork aggravated the pain. There was no personal or family history of headache, and no secondary causes were identified. On the basis of this history, she met the criteria for NDPH.
The patient’s participation in soccer over the years led to sports-related injuries, and on multiple occasions she had fallen on her chest while diving for a ball, including the day her headache started. She had not directly injured her neck at the time, and she denied head butting the soccer ball. The patient did report trauma to the midthoracic spine from a wave while standing in the ocean, which occurred after the diagnosis of NDPH. She also had a history of bilateral Osgood-Schlatter disease and numerous athletic injuries to the knees and ankles. Multiple treatment approaches had been attempted during the 2 years before the current presentation, including amitriptyline, topiramate, and sumatriptan, as well as Chinese medicine, homeopathy, and acupuncture.

Results of a general physical examination, including her body habitus, were normal. A neurologic examination revealed bilateral forehead allodynia, normal bulk and tone with full strength, normal coordination, proprioception, vibration and pin sensation, and normal station and gait. Her reflexes were brisk (3/5) but symmetric with flexor plantar reflexes.

Results of an osteopathic structural examination revealed that the occipitoatlantal joint was severely extended, rotated right, and sidebent left; atlantoaxial joint was rotated right and sidebent left; second cervical vertebra was extended, rotated left, and sidebent left (with hypomobility of the occipitoatlantoaxial joints); and left frontal bone had external rotation with dural compression. Severe limitation was found in the motion of the left temporal bone, left maxilla, vomer, and mandible, but a definitive strain pattern was not recorded. Restriction cephalad was demonstrated in both the anterior and deep anterior thoracic fascia; T4 was flexed, rotated right, and sidebent right; L5 was rotated left and sidebent right; left anterior innominate was rotated left; left unilateral sacral was extended; and left leg had external rotation. Results of cranial magnetic resonance imaging were normal.

After this osteopathic structural examination, a course of OMT was started, but the patient could not tolerate techniques applied to the head. Techniques were therefore directed at the extracranial areas of somatic dysfunction. The OMT techniques used included osteopathic cranial manipulative medicine, balanced ligamentous tension, myofascial release, muscle energy, and muscle inhibition. After 3 weekly sessions, her phonophobia was more tolerable, but her headache did not change. After 5 OMT sessions, she had 1 to 2 days of level 7 to 10 pain, with more variability in the pain. After 7 treatment sessions, she no longer experienced exacerbations between 7 and 10 pain, and her pain level decreased to between 5 and 10 on most days. All areas of somatic dysfunction resolved with the exception of the head and neck.

The patient could not tolerate treatments to the head because of the severe forehead allodynia. Occipitotemporal hold was not sufficient to manage the frontal bone and dural dysfunction. Instead she required direct manipulation on the frontal bone. Beginning with treatment session 16, her phonophobia continued to improve, so she was able to resume school activities, sports, and choir. Treatments were then directed at the atlantoaxial junction, but this area was treatment resistant. Her headaches remained stable, between 5 and 10 pain, with exacerbations between 6 and 10 twice per month.

She eventually permitted cranial manipulation at the occiput at treatment session 33. She continued to note improvement in her phonophobia and endurance and did not report exacerbations of pain despite an unchanged cranial and cervical strain pattern. Her headache continued to range between a pain level of 5 and 6. She subsequently allowed cranial manipulation to the whole head at treatment session 37 and noted the largest single improvement in her symptoms after 1 treatment since starting OMT. Her headache completely resolved after the third application of OMT to her whole head. Normal range of motion of the occipitoatlantoaxial joints, neutral occiput, C2, and frontal bone were observed. The patient received a total of 40 OMT sessions over 1.5 years. Treatments were administered weekly when possible, but gaps occurred.
Discussion

The pathophysiologic mechanism of and effective managements for NDPH remain elusive. Spontaneous resolution can occur, but there is wide variability in patient experience and duration of symptoms, which can last from months to decades.1,6

The head pain associated with NDPH occurs in the trigeminal nerve distribution that innervates the dura. The dura may be a primary cause of pain in headache or a site of referred pain. Referred pain from the cervical spine is felt in the cranial trigeminal nerve distribution, which is the likely mechanism in cervicogenic headache.7 The trigeminal nerve caudalis transmits pain information from the cervical spine to the trigeminal ganglion, which sends branches to the tentorium cerebelli and middle cranial fossa dura as well as the 3 main trigeminal nerves.

The fact that the patient’s pain did not resolve until her frontal bone and dural somatic dysfunction were addressed supports the dura as being the main area of injury in NDPH. This injury may have occurred from trauma at the C2 region acquired while repeatedly diving for a soccer ball and landing on her chest. However, the patient had a compression pattern from an unclear origin near the frontal bone. The patient denied head butting in soccer, but such activity could have explained the frontal compression pattern. The frontal bone, occiput, and C2 interrelate via the reciprocal tension membrane formed by the falx cerebri, falx tentorium, and the dural attachment at C2. Release of the frontal bone in turn released both the dural somatic dysfunction and the occipitoatlantoaxial dysfunction, which excludes injury at C2 from being the main source of the headache. It may be that unrecognized repeated head trauma from playing sports caused the cervical dysfunction.

Interestingly, this patient’s symptoms improved but did not resolve with OMT techniques targeting extracranial areas of dysfunction, implying a contribution from musculoskeletal lesions distal to the pain site. Further, we had to target these areas before the head, supporting the idea that the somatic dysfunction of the extracranial regions influenced that of the cranium. This relationship is likely due to fascial continuity and the effect of the myodural bridge, a communication between the epidural spaces, suboccipital muscles, and dura at C2.8 The soccer injuries probably played a secondary role. Substernal fascial strain was observed. This fascia is continuous with the pretracheal fascia, which attaches to the occiput. It is noteworthy that some of these areas of somatic dysfunction were sites of injuries received years before as well as after the onset of her NDPH, which may account for the long duration of treatment needed. These earlier traumas may have sensitized her system for NDPH.

Although the literature describes hypermobility as possibly being associated with NDPH, this patient had restricted mobility at the occipito-atlanto-axial joints. The role of somatic dysfunction distal to the primary pain site may have indicated a referred pain syndrome, but it does not account for the treatment refractory nature of the somatic dysfunction at this site and the rapid resolution of the headache with correction of the frontal bone and dural somatic dysfunction. Whether the pain from somatic dysfunction is primary or referred, this case suggests a biomechanical component to NDPH.

Conclusion

The impact of OMT on NDPH is unclear. In this case report, spontaneous resolution cannot be excluded. However, when viewed in the context of studies that have shown the influence of joint hypermobility, myotomy, and the role of the myodural bridge on headache, the current case report supports a musculoskeletal component to head pain.5,6 It is not known whether NDPH is a primary or secondary problem, but this case suggests both as mechanisms. The biomechanical model of headache and the role manual treatments have in management, particularly for NDPH, require further exploration to determine whether they are a promising approach to headache.
References


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