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Diagnosis, Management, and Return to Sport of a 16-Year-Old Patient With a Chiari I Malformation: A Case Report and Literature Review

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This case report discusses a 16-year-old female volleyball, basketball, and track and field athlete who was diagnosed with a Chiari I malformation after a concussion. Surgical decompression was recommended and performed 3 months after her initial diagnosis. This patient presented unique challenges due to her age, desire to return to sport, and lack of access to medical care due to living in a rural area. Few evidence-based best-practice recommendations are available for the management and return to sport of patients with Chiari I malformation, particularly for those who have undergone surgical decompression. In this case study, we address the treatment and return-to-sport process for the patient and provide a comprehensive review of the published literature on patients attempting to return to sport after a diagnosis of Chiari I malformation. In addition, we explore the value of an athletic trainer in reconciling various barriers in management and return to sport evident in this case and the reviewed literature.

Key Words: brain, congenital deformity, return to play

Key Points

- Chiari I malformations are a rare cause of neurologic symptoms in athletes and are often diagnosed incidentally after other head injuries.
- No consensus exists on the most appropriate management of athletes with Chiari I malformations, particularly for those returning to sport after surgical decompression.
- Athletic trainers should recognize the potential for symptom recurrence after return to sport following decompression surgery and work with the physician and surgeon to develop an appropriate care plan.

Arnold-Chiari malformations are a spectrum of congenital deformities of the hindbrain with 4 classifications and various comorbidities.[1–5] *Chiari I malformations* (CIMs) are the most common of the 4 classifications and are defined as having a >5-mm caudal herniation of the cerebellar tonsils below the foramen magnum.[2] The exact cause of CIMs remains uncertain, although researchers have proposed several theories, including (1) interrupted cranial development, (2) irregular cerebrospinal fluid (CSF) circulation causing altered intracranial pressure, and (3) pressure on the brainstem and cerebellum caused by hydrocephalus.[3,5,6] The structural alterations due to CIMs create 2 distinct pathophysiological changes that result in symptom development. These mechanisms are direct compression of the brainstem and upper cervical spinal cord and obstruction of the CSF circulation, which can result in hydrocephalus or syringomyelia.[2,4,6] Suboccipital headaches are the most common symptom experienced by patients with CIM, found in 80% of cases, and are often precipitated by Valsalva-like actions such as coughing, sneezing, laughing, straining, vigorous lifting, and strenuous physical activity.[2] Patients may also present with ocular, otoneurologic, lower cranial nerve, myelopathic, or cerebellar symptoms.[1–8]

Considered a rare condition, CIMs have an incidence ranging from 0.56% to 4%.[1,5,9,10] They are often referred to as the "adult" form of Chiari malformations because symptoms typically do not occur until the third or fourth decades of life.[5,9] Aitken et al[10] reported that 14% to 30% of adult patients and 37% to 57% of pediatric patients presented asymptomatically. This may reflect the fact that syringomyelia is found in 59% to 76% of adult patients and only 14% to 58% of pediatric patients. In addition, only 21% of asymptomatic pediatric patients will eventually develop symptoms, usually headaches and neck pain.[6,10] Meadows et al[9] retrospectively reviewed all magnetic resonance imaging (MRI) scans of the brain and cervical spine taken over a 43-month period in a single hospital. The patients were aged 1 to 65 years; of the 22 591 MRI scans, the researchers identified only 175 CIMs, and 25 patients were considered asymptomatic at the time of the scan. Aitken et al[10] retrospectively reviewed all brain and cervical spine MRIs over a 24-month period from multiple hospitals of patients ages 1 to 20 years. Of the 5248 scans, the authors found Chiari malformations on only 51.

Whereas CIM is a rare diagnosis, case reports of athletic patients with CIMs have become more prevalent. In athletic populations, CIMs are often incidentally diagnosed through MRI ordered after the athlete incurs a head or neck injury, such as a concussion. As outlined by Bonow et al,[11] incidental MRI findings after concussions are not uncommon. In a review of 427 MRIs of pediatric patients who sustained concussions, 52 incidental abnormalities were found, 8 of which were CIMs.[11] Concurrently, the authors of various case reports have described the incidental diagnosis of CIMs (Table 1), which suggests inherent concerns about the risk of sport participation for these patients. However, consensus is lacking on best practices due to the absence of current, evidence-based research to support firm recommendations for sport participation for patients with CIM. This leads to clinicians relying on expert opinion and case reports in the management of sport participation. Therefore, the purpose of our article is 2-fold. First, we present the unique challenges faced by an adolescent athlete who was diagnosed with a CIM, was treated surgically, and eventually was able to return to sport (RTS). Second, we comprehensively summarize the existing case studies on young athletes to compare and contrast this case with others, determine how health care professionals are managing and making RTS decisions for athletes after CIM diagnosis, identify barriers affecting the management and RTS of young athletes with CIM, and provide recommendations to address those barriers.

CASE PRESENTATION

The patient was a 16-year-old female high school volleyball, basketball, and track and field athlete. In July 2018, she had the acute onset of headaches, dizziness, and left-sided neck pain that persisted for 7 days. She was engaged in rigorous volleyball practices at the time, but no clear injury was linked to the onset of her symptoms. Her headaches became more intermittent and lasted about 1 hour before resolution. The headaches would often wake her up at night, and she reported headaches upon waking most mornings. She also described sharp pain radiating from her left occiput to her left parietal and frontal areas. Her symptoms were frequently provoked by participating in physical activity and were slightly palliated at rest.

During a volleyball tournament in mid-August 2018, she sustained a concussion that was diagnosed by her rural outreach athletic trainer (AT). At this time, the AT instructed the patient to cease participation in athletics and physical activity. Her preexisting headache, neck pain, and dizziness all worsened after her concussion, and the AT recommended she be seen by her primary care provider (PCP). During her follow-up appointment with the PCP on August 31, 2018, she reported new symptoms, including tremors in her hands and both near- and full-syncopal events when rising from seated or lying positions. She also noted numbness and tingling in her arms, hands, and legs that seemed to be positional and were alleviated when she changed positions. She believed the symptoms had gotten worse since her first appointment. Her PCP ordered a brain MRI scan, which revealed a CIM with significant left cerebellar tonsillar herniation to the upper aspect of C2 and slightly less right cerebellar tonsillar herniation to C1 (23–25 mm).

After the diagnosis, the athlete ceased participation in athletic physical activity. She was referred to a neurosurgeon for surgical evaluation. Her initial neurologic examination was completely normal: no evidence of nystagmus, normal gag reflex, full strength and normal sensation throughout her extremities, normal reflexes with no pathologic reflexes, and no cerebellar signs. Given her presenting symptoms and the severity of the MRI findings (>20-mm cerebellar tonsillar herniation), surgical intervention was recommended. On October 24, 2018, she underwent successful suboccipital craniectomy, C1 laminectomy, and expansile duraplasty for CIM decompression. Her postoperative course was uneventful. She was mobilized with physical therapy on the first day after surgery and was discharged home 3 days after surgery with a normal neurologic examination. She was given strict instructions to avoid sports, strenuous activity, swimming, and heavy lifting (>10 lb [4.5 kg]). She was permitted to return to school 2 weeks after her discharge, starting with half days on November 12 and progressing to full days as tolerated 1 week later.

On January 28, 2019, at her second neurosurgery postoperative appointment, she reported that most of her preoperative symptoms were resolved. However, she continued to experience near-syncopal events that were similar in frequency to her preoperative events, including a full-syncopal event on November 22, 2018. The neurosurgeon felt these events were unrelated to her Chiari decompression surgery and were more likely a result of orthostatic hypotension. Fluid hydration was encouraged, and if the events persisted, a referral to cardiology was recommended. Her neurosurgeon cleared her for a slow but eventual full RTS and advised her to monitor her symptoms closely. The following week, she began her RTS by going to her gym 3 days per week and performing low- to moderate-intensity cardiovascular exercise and shooting basketballs. For the next 3 weeks, she attended basketball open gym, which consisted of 5 days a week of moderate- to high-intensity cardiovascular exercise for 2 hours per day. She then joined the track team and began maximum-effort participation. On April 6, she described recurrent headaches and tremors that occurred during and after vigorous physical activity, primarily after track practice. The headaches were as severe as her preoperative headaches but in a different location, and the tremors were the same as before surgery. These symptoms went away with rest. On April 26, 2019, she had a follow-up appointment with her neurosurgeon and new MRI to discuss the recurrence of symptoms. The MRI showed good decompression of the CIM with no concerning changes at the surgical site or new abnormalities. Her neurologic examination remained completely normal with no pathologic findings. With attention to adequate fluid hydration, she was no longer having syncopal episodes. Her neurosurgeon believed the recurrence of symptoms was unrelated to her CIM and was more likely related to overexertion during exercise. The patient remained cleared for full RTS but was instructed to advance her activities a bit more slowly, to rest if symptoms recurred, and to not pursue a full RTS until symptoms completely subsided. She was able to slowly increase her participation in vigorous physical activities and eventually these recurrent symptoms resolved. Her frequency of follow-up appointments with

Table 1. Case Report Summary

Report	Age, y	Sex	Sport(s)	Presentation	Symptomatic Before Diagnosis?	Incidental Diagnosis?	Surgical Treatment?	Return to Sport?	Symptom Recurrence?
Asymptomatic patients at diagnosis									
Callaway et al (1996)[18]	8	M	American football	Neck-axial and -flexion injury in game → bilateral anterior thigh tingling	No	Yes	Yes: Suboccipital craniotomy and C1 laminectomy	No: Ended football participation and proceeded with surgery after various MD consultations	No
Kurup et al (2005)[19]	17	M	American football	Neck-flexion injury in game, transported to emergency department for residual neck pain and neurologic symptoms	No	Yes	No	Yes: Returned against MD's recommendation	No
Hunt and Amato (2007)[20]	18	F	Volleyball	Concussion and possible post-concussion syndrome, failed to improve on conservative management x 10 wk	No	Yes	No	Yes: Returned after MD clearance as long as she was asymptomatic	No
Kirschen and Illes (2014)[15]	17	M	American football	No complaints at mandatory preparticipation neurologic evaluation	No	Yes	No	Yes: Returned after MD clearance understanding and accepting risks of participating with asymptomatic CIM	No
Symptomatic patients at diagnosis									
Lewis et al (2008)[21]	22	M	American football	Progressive left upper arm pain and weakness exacerbated by contact drills in practice and upper extremity changes in temperature sensation	Yes: Upper arm weakness and pain during spring football camp 18 mo prior that resolved after he ceased football until symptoms subsided	No	No	No: Ended football participation after MD consultation	No
Lewis et al (2008)[21]	18	M	Baseball	Left arm and shoulder fatigue, loss of accuracy and velocity	Yes: Sharp pain radiating from rib cage up left arm and shoulder x 4 mo, left arm and shoulder changes in pain and temperature sensation x 3 mo	No	Yes: Suboccipital craniotomy and C1 laminectomy with duraplasty	Yes: Returned with MD clearance	No
Walk (2008)[22]	20	M	American football	Right knee pain, ↓ sensation in left foot, + Babinski sign, difficulty urinating, ↓ balance, headaches; immediately taken to emergency department	Yes: Symptoms ↑ after football game 3 mo prior	No	Yes: Suboccipital craniotomy and C1 laminectomy	Not available	No
Harrell and Barootes (2010)[23]	19	M	American football	Severe frontal headache, mild photophobia and nausea	Yes: Symptoms began 3 d prior	No	No	Yes: Returned with MD clearance	No
Starnes et al (2015)[24]	15	M	American football	Concussion-like symptoms in game 3 d prior with no concussion mechanism of injury; removed from play and evaluated	Yes: Previous CIM diagnosis and surgical treatment; was asymptomatic until this incident	No	Yes: When initially diagnosed	Yes: Diagnosed with concussion and cleared after completing return-to-sport protocol	No: After this incident of symptom recurrence, he underwent conservative management and became asymptomatic again.
Zhang et al (2016)[25]	20	M	Lacrosse	Left shoulder pain and weakness, left trapezius and deltoid atrophy	Yes: Symptoms began 6 y prior after shoulder dislocation and surgery for shoulder instability; became progressively worse	No	Yes: Suboccipital craniotomy and C1-C2 laminectomy with duraplasty	Yes: Returned with MD clearance	No
Ando et al (2017)[1]	18	M	American football	Concussion during practice, transported to emergency department	Yes: Headaches x 2 y	Yes	Yes: Posterior fossa decompression and external durotomy	No: Ended football participation due to symptom persistence and recurrence after various MD consultations	Yes: Symptoms returned by 18 mo postsurgery
Kennedy et al (2019)[26]	15	F	Band, gymnastics	Nonspecific, right-sided neck pain	Yes: Neck pain x 2 y	No	Yes: Posterior fossa decompression and C1 laminectomy	Yes: Returned to participation in band but not gymnastics	No
Kennedy et al (2019)[26]	15	F	Cheer	Right anterolateral neck pain	Yes: Neck pain x 4 mo	No	Yes: Posterior fossa decompression and C1 laminectomy	No: Ended participation	No

Abbreviations: CIM, Chiari I malformation; F, female; M, male; MD, doctor of medicine.

her neurosurgeon was changed to once annually, and on starting college in August 2020, she remained symptom free with no neurosurgical follow-up required.

DISCUSSION

In the competitive athlete, CIMs are a rare but possible diagnosis that can present with a wide variety of symptoms, making them challenging to diagnose by symptoms alone. Some of the most common symptoms of CIM, including headaches, neck pain, and paresthesias, are nonspecific and are also commonly associated with many other conditions, such as concussion, anxiety, and migraine syndromes.[6,7,10] Specific headache patterns, including headaches that only occur with exertion, coughing, or straining; suboccipital location of headaches; new onset or increasing frequency of headaches; or headaches accompanied by other neurologic symptoms all warrant referral for a more complete diagnostic work-up.[12,13] In addition, any abnormal neurologic examination findings, including altered reflexes or altered sensory or motor function that does not correspond with a specific mechanism of injury, ataxia, cranial nerve dysfunction, nystagmus, or unexplained syncope, warrant referral and disqualification until the athlete is evaluated and cleared by a physician.[7] Imaging, specifically head and cervical spine MRI, is the study ordered most frequently to look for structural abnormalities such as CIM. Once a CIM is diagnosed, the decision regarding surgical intervention is based on the neurologic examination, imaging findings, and symptom severity. Chiari I malformation is generally treated with surgery in patients who have an abnormal neurologic examination, MRI findings of structural compression of the brainstem with evidence of impaired CSF circulation, hydrocephalus, syringomyelia, or clinical symptoms significantly affecting the quality of life.[6,7]

The primary concern for sport participation in patients with CIM, whether they are treated conservatively or surgically, is the potential risk of sustaining traumatic or catastrophic head or spinal injuries and recurrence of symptoms.[14,15] Meehan et al[14] and Kirschen et al[15] stated that no evidence indicated that athletes with CIM were at increased risk of sustaining traumatic or catastrophic head or spinal injuries. However, Tator et al[16] and Spencer et al[17] suggested that patients with CIM had an increased chance of sustaining concussions and postconcussion syndrome, which could induce new or worsen preexisting CIM symptoms. As a result of the mixed literature findings on RTS risks, clinicians and researchers have been unable to draw a definitive conclusion regarding the risk of RTS for patients with CIM. Although case reports and expert opinions are not as definitive as evidence-based practice, many clinicians must rely on them for RTS decisions. The various case reports provided information about athletic patients with CIM as well as the barriers encountered and strategies used in diagnosis, management, and RTS. The overall consensus from the case reports summarized in Table 1 was that asymptomatic patients diagnosed incidentally and who sustained concussions or head or neck injuries generally responded well to conservative management and were able to RTS.[15,18–20] Symptomatic patients generally underwent surgical treatment; approximately half were able to RTS and then responded positively to conservative management if symptoms recurred. Most cases (7/12) involved male American football players, which could be due to the increased risk of concussions and head or neck injuries in the sport, thereby leading to imaging and diagnosis.[1,15,17–19,21–26] Most researchers[1,23] also reported that patients were cleared to return to activity by their physician and those who did not RTS were often given autonomy in their RTS decision.

Clinicians have varied opinions on whether CIMs are an absolute or relative contraindication for participation in sports.[14] Kirschen et al[15] found that 18% to 36% of physicians considered a diagnosed Chiari malformation a contraindication for participation in sport and an indication for surgical intervention. An asymptomatic adult with a normal neurologic examination may be given autonomy to decide whether to RTS as long as the patient understands and agrees to the risks. However, for minor and collegiate athletes, a team approach involving the physician, parent(s), team medical providers, and athlete is necessary, along with a plan for very close monitoring by the AT if the individual does RTS.[14,15,27] Most clinicians believed the benefits of participation in sport outweighed the risks for asymptomatic CIM patients, barring the onset of symptoms or development of an abnormal neurologic examination.[14,15,19] The specific MRI findings also played a role in this conversation, with significant cerebellar tonsillar herniation (>5 mm) being a more at-risk finding than mild cerebellar tonsillar ectopia (<5 mm). Some asymptomatic patients may request prophylactic surgery, but this is strongly discouraged due to the risk of surgical complications and the possibility of poor surgical outcomes.[6,14,15]

Postsurgery patients with CIM who RTS also face risks that can include symptom recurrence, postoperative complications, and an increased risk of head or neck injury due to the surgical bone removal and weakened neck musculature after surgery. Certain sports that have a high risk of head or neck injury (ie, football, wrestling, rugby, and gymnastics) likely pose a greater risk to postsurgical patients than do other, less aggressive and noncontact sports.[6] In a retrospective review of 256 pediatric cases, McGirt et al[28] observed that 22% of patients had persistent or recurring symptoms and 7% of those required surgical revision. Headaches were 70% more likely to persist or recur than any other symptom. The chance of symptom recurrence increased by 15% for each year headaches grew worse before surgery. In addition, preoperative cerebellar tonsillar herniation down to the C2 lamina doubled the chance of needing a surgical revision. Arnautovic et al[29] conducted a similar retrospective review of 145 (30% pediatric, 27% adult, 43% unknown) postoperative patients and determined that 75% improved, 26% had persistent or recurrent symptoms, and 41% had postsurgical complications including CSF leak, pseudomeningocele formation, aseptic meningitis, wound infection, meningitis, or new neurologic deficit.

Based on the available data, the diagnosis and surgical management of a CIM is not an absolute contraindication for participation in sports, but many patients do experience some symptom recurrence after surgery. The athlete's neurologic examination and specific symptoms are the most important factors in determining the next steps in management. Certain symptoms and examination findings are critical for a health care professional (including an AT) to recognize in order to guide management regarding referral to a specialist versus continuation of an RTS plan.

Table 2. "Red-Flag" Symptoms and Associated Signs Necessitating Disqualification From Sport and Referral for Medical Evaluation[a]

System	Symptom(s)	Signs
Central nervous system		
	Nausea with vomiting	Confusion
		Altered mental status
	Headache	Short headache duration
	• Acute onset associated with injury	Frequent use of over-the-counter medications
	• Exertional and resolves at rest	
	• Increasing frequency or severity	
	• Preventing school attendance	
	Vision (acute onset)	Nystagmus
	• Blurry vision	Visual acuity change
	• Double vision	Dysconjugate gaze
	Hearing	Altered hearing
	• Tinnitus (ringing in ears)	
	Balance or vestibular	Difficulty with tandem gait
	• Dizziness or balance difficulty	Difficulty with single- or double-legged stance
		Gait ataxia
Peripheral nervous system		
	Sensory	New sensory dysfunction (even if transient)
	• Numbness/tingling or "stinger"	Any new sensory asymmetry
	Motor or reflex	New weakness (even if transient)
	• Weakness or "stinger"	Any new motor asymmetry
		Altered deep tendon reflex/hyperreflexia
		+ Babinski sign
		Monoparesis, hemiparesis, or quadriparesis requires emergent evaluation
Cervical spine or neck	Neck pain	Torticollis
	• Acute onset associated with injury	Swelling at surgical site
	• Increasing frequency or severity	

[a] For any athlete without a known diagnosis, with a diagnosis of nonoperated Chiari I malformation, or returning to sport after Chiari I malformation surgery.

Suggested "red-flag" signs and symptoms for ATs (and other health care providers) that warrant disqualification from participation and referral to the patient's PCP, neurologist, or neurosurgeon for further examination and recommendation regarding continued sport participation are provided in Table 2. Whereas most referrals from the AT will be to the athlete's PCP or neurosurgeon, it is important to recognize that other medical providers may become involved in the athlete's care. If no additional surgical concerns are identified, the PCP may refer the athlete for a neurologic or mental health evaluation to further assess persistent symptoms for a neurologic or possibly psychosomatic cause. Thus, having access to appropriate medical care is critical for patients who elect to RTS. Patients in larger metropolitan areas likely have quick access to their PCP or neurosurgeon, but rural patients face significant barriers in receiving specialized care. Neurosurgeons encounter considerable challenges when working with rural patients, due to long wait times for nonemergent appointments, increased travel times, and delayed appointments, as well as diagnoses that are sometimes challenging.[30] Our patient lived roughly 200 miles from her neurosurgeon, and her closest opportunity for medical care was her PCP. In addition, she had no access to a provider with expertise in sports medicine and RTS and was, therefore, left with minimal support as she navigated her RTS after surgery. The patient would have benefited from access to a sports medicine clinician trained in RTS, such as an AT, to assist in the management and monitoring of her symptoms.

Athletic trainers provide evaluation, diagnosis, prevention, rehabilitation, and treatment of acute, chronic, and emergent injuries and medical conditions. In secondary school settings, ATs supply these services to pediatric athletic patient populations and thus must be well versed in RTS decision making.[31] Furthermore, by monitoring and tracking symptoms, ATs can facilitate referral for atypical and red-flag signs and symptoms and oversee appropriate additional conservative management. Athletic trainers deliver high-quality management of concussions and other injuries and are particularly helpful in the routine care of patients who may present with chronic or comorbid conditions. More than 92% of ATs agreed with physicians regarding the diagnosis of patients with concussions and mild traumatic brain injuries, and concussion management was enhanced when ATs were readily available.[22,32,33] Thus, an AT may be a critical partner in the care of athletes diagnosed with CIM, particularly to monitor for symptom development or recurrence and differentiate between CIM symptoms and potential new head injuries such as concussions.

CLINICAL BOTTOM LINE

The patient in this case study was diagnosed with a CIM after sustaining a concussion; she underwent successful decompression surgery but experienced some symptom recurrence after her RTS. Few evidence-based best-practice recommendations guide the management and RTS for patients with CIM. Clinicians base most of their decisions on consensus from case reports and expert opinions. Approximately 50% of CIM patients treated with surgical decompression experience symptom recurrence after RTS, which suggests that they may need close monitoring and careful evaluation if or when symptoms occur. For many patients with CIM involved in sport, ATs may be the first health care providers to identify the onset of symptoms, perform a neurologic examination, and refer the patient to another provider for evaluation. After diagnosis, ATs are often the most convenient source of monitoring for the onset or worsening of symptoms during sport participation and are the first responders in the case of traumatic injuries sustained in sport. We presented a unique perspective on a patient with CIM diagnosed incidentally via MRI after a concussion. This case also highlights the many barriers faced in the diagnosis, management, and RTS of patients with CIM that an AT may be able to resolve and a unique opportunity for ATs to advance the best-practice recommendations for RTS and overall management of these patients.

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