Sport-related structural brain injury associated with arachnoid cysts: a systematic review and quantitative analysis

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OBJECTIVE Arachnoid cysts (ACs) are congenital lesions bordered by an arachnoid membrane. Researchers have postulated that individuals with an AC demonstrate a higher rate of structural brain injury after trauma. Given the potential neurological consequences of a structural brain injury requiring neurosurgical intervention, the authors sought to perform a systematic review of sport-related structural-brain injury associated with ACs with a corresponding quantitative analysis.

METHODS Titles and abstracts were searched systematically across the following databases: PubMed, Embase, Cinahl, and PsycINFO. The review was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement. Peer-reviewed case reports, case series, or observational studies that reported a structural brain injury due to a sport or recreational activity (hereafter referred to as sport-related) with an associated AC were included. Patients were excluded if they did not have an AC, suffered a concussion without structural brain injury, or sustained the injury during a non-sport-related activity (e.g., fall, motor vehicle collision). Descriptive statistical analysis and time to presentation data were summarized. Univariate logistic regression models to assess predictors of neurological deficit, open craniotomy, and cystoperitoneal shunt were completed.

RESULTS After an initial search of 994 original articles, 52 studies were found that reported 65 cases of sport-related structural brain injury associated with an AC. The median age at presentation was 16 years (range 4–75 years). Headache was the most common presenting symptom (98%), followed by nausea and vomiting in 49%. Thirteen patients (21%) presented with a neurological deficit, most commonly hemiparesis. Open craniotomy was the most common form of treatment (49%). Bur holes and cyst fenestration were performed in 29 (45%) and 31 (48%) patients, respectively. Seven patients (11%) received a cystoperitoneal shunt. Four cases reported medical management only without any surgical intervention. No significant predictors were found for neurological deficit or open craniotomy. In the univariate model predicting the need for a cystoperitoneal shunt, the odds of receiving a shunt decreased as age increased (p = 0.004, OR 0.62 [95% CI 0.45–0.86]) and with male sex (p = 0.036, OR 0.15 [95% CI 0.03–0.88]).

CONCLUSIONS This systematic review yielded 65 cases of sport-related structural brain injury associated with ACs. The majority of patients presented with chronic symptoms, and recovery was reported generally to be good. Although the review is subject to publication bias, the authors do not find at present that there is contraindication for patients with an AC to participate in sports, although parents and children should be counseled appropriately. Further studies are necessary to better evaluate AC characteristics that could pose a higher risk of adverse events after trauma.

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KEY WORDS sports concussion; traumatic brain injury; arachnoid cyst; subdural hemorrhage; hygroma; return to play

Sport-related concussion (SRC) has emerged as a public health problem, affecting athletes of all ages and participation levels.11,35,43 While SRC by definition includes normal imaging results, a minority of sport-related head injuries result in structural brain injury.3,18,19,84,85 These cases are often neurosurgical emergencies and can result in permanent neurological deficit and/or death. Recent literature has postulated that athletes with arachnoid cysts (ACs) demonstrate a higher rate of structural brain injury after trauma, specifically subdural hematoma (SDH) or hygroma.37,44,79

ACs are congenital lesions bordered by an arachnoid
Cysts are filled with CSF, occur in 0.7% to 1.7% of the general population,66,76 and are found most commonly on the left side with a 3:1 male to female predilection.37,78 Several reviews have discussed the risk of SDH or hygroma associated with ACs after head trauma.37,44,79 The underlying mechanism for these structural injuries is unknown, but theories propose that 1) vessels without support of the cyst wall are prone to rupture,17,66 2) vessels within the wall can rupture due to decreased compliance,55 or 3) a slit-valve mechanism is created, leading to increased pressure within the AC and vessel rupture.64 A select few reports have addressed the issue of return to sport with an AC, with or without structural injury.21,44

Given the uncertainty surrounding structural brain injuries in athletes with an AC and the experiences at our own neurosurgical department and sports concussion center, we sought to review the literature in this area. The goals of this systematic review were to identify all cases of sport-related structural-brain injury associated with ACs and to perform a quantitative analysis in this unique patient population. Both objectives represent an overarching goal to better characterize, prevent complications, and counsel about return to play in athletes with an AC.

Methods

Published titles and abstracts in the English language were searched systematically across the following electronic databases: PubMed, Embase, CINAHL, and PsycINFO. The search terms included MeSH terms and keywords. Search words included the following: arachnoid cyst, sport-related concussion, subdural hematoma, subdural hygroma, sports injuries, and athletic injuries. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses45 were adapted to review a heterogeneous collection of literature. A flow diagram of the included studies is seen in Fig. 1. Example search syntax from PubMed is noted below:


Inclusion criteria consisted of the following: a case report, case series, or observational study of a patient with a structural brain injury due to a sport or recreational activity with an associated AC. Structural brain injury was defined most commonly as an acute or chronic SDH or subdural hygroma, in addition to less common injuries, such as epidural hematoma (EDH), subarachnoid hemorrhage (SAH), intraparenchymal hemorrhage (IPH), and skull fracture. “Sport-related” was defined according to definitions in earlier literature,84 specifically any endeavor in which an individual was performing an athletic activity alone or in competition against an opposing person or team. Patients were excluded if they did not have an AC, suffered a concussion without structural brain injury, or sustained the injury during a non–sport-related activity (e.g., fall, motor vehicle collision). While the question of whether patients with an AC are at increased risk of structural brain injury is an important one, incidence data cannot be calculated from the current systematic review. Rather, the question of whether athletes with an AC can return to play will be addressed.

After the initial search was completed, as described in Fig. 1, a separate review of all references of included studies was completed to ensure no cases were missed. All references of included studies were systematically reviewed. Of the 980 references from included papers, 169 full-text sources were searched for eligibility. All but 3 were excluded for the following reasons: unrelated (n = 97), unspecified or not a sport-related trauma (n = 8), or no trauma mentioned with AC complication (n = 61). The additional 3 sources were included in the final 52 articles.

Statistical Analysis

A descriptive statistical analysis was performed. Due to the limited sample size, a normal distribution was not assumed, and all data were considered to be nonparametric. Medians, interquartile ranges (IQR), and ranges were provided for continuous variables. Univariate logistic regression was performed to predict certain presentations or outcomes. A priori exposure variables included age, sex, days to presentation, presence of midline shift, and presence of a SDH. Multivariate analysis was not used due to limited sample and concern for an unstable model. Time to presentation data were represented with a Kaplan-Meier time to failure plot.29 All statistical analyses were performed in STATA (version 14, StataCorp LP).

Results

Our review yielded 52 studies reporting 65 cases of sport-related structural brain injury associated with an AC. The median year of publication was 2005, ranging from 1958 to 2015 (Fig. 2). The median age at presentation was 16 years (4–75 years) (Fig. 3), and 56 patients (86%) were male. Age was not mentioned in 2 studies, whereas sex was mentioned in all.

Presentation

Presentation data are summarized in Table 1. The median time from either injury or symptom onset to discovery of the structural injury was 25 days, ranging from 1 to 168 days. Of all 65 patients, symptom endorsement was mentioned in 61 (94%) of studies. Headache was the most common symptom, presenting in all but 1 patient, followed by nausea and vomiting in 49%. Thirteen patients (21%) presented with a neurological deficit, most commonly hemiparesis. The range of sporting activities across...
all 52 studies is detailed in Table 2. Team sports were involved in 28 cases (43%).

**Imaging and Treatment**

Details regarding imaging and treatment are illustrated in Table 3. A CT scan and/or MRI of the head was obtained in all 65 cases. SDH was the most common structural injury (72%) followed by hygroma (23%), EDH (2%), and IPH (2%). The most common imaging finding of the structural abnormality (SDH or hygroma) on CT was an iso-/hypodense lesion signifying a chronic injury (76%). Consistent with prior literature, 75% of patients harbored left-sided ACs. Due to the heterogeneous terminology and lack of complete CT/MRI studies, middle fossa and sylvian locations were combined and represented 98% of cases. Skull thinning adjacent to the cyst was seen in 60% of cases and often mentioned as a clue for AC presence when visualization was difficult, most often in the case of intracystic hemorrhage or an emergent, life-saving situation.

Four cases (6%) reported medical management only without any surgical intervention; in 1 of these cases the family was offered surgery but refused. Two cases were hygromas that were followed with serial imaging and were noted to decrease over time, and one was an isolated 2.3-cm IPH on the opposite side of the AC. Of the remaining 61 patients (94%), open craniotomy alone was performed.
in 29 patients (45%) and was the most common surgical procedure, followed by bur holes alone in 26 patients (41%); 3 patients (5%) underwent both. During the open craniotomy or bur hole, cyst fenestration was performed in 31 patients (48%). Seven patients (11%) received a cystoperitoneal shunt. Five patients (8%) underwent more than 1 operation. An endoscope was used in 2 cases (3%). Postoperative outcome was mentioned in 36 cases (55%). Each of the 65 cases is summarized in Table 4.

Three univariate models were conducted in an attempt to predict 1) a neurological deficit, 2) need for open craniotomy, and 3) need for a cystoperitoneal shunt (Table 5). The predictive factors in each model were determined a priori and included age, sex, days to presentation, midline shift, and presence of an SDH. No significant predictors were found for neurological deficit or open craniotomy. In the model predicting the need for a cystoperitoneal shunt, the odds of receiving a shunt decreased as age increased ($p = 0.004, \text{OR} 0.62 [95\% \text{ CI} 0.45–0.86]$) and with male sex ($p = 0.036, \text{OR} 0.15 [95\% \text{ CI} 0.03–0.88]$).

**Discussion**

While posttraumatic SRC in general has been well studied, less is known about structural brain injuries in sports. ACs are congenital malformations with the potential to hemorrhage after relatively minor head injury. The goal of this systematic review was to summarize all cases of sport-related structural brain injury associated with ACs and to provide a quantitative analysis. A total of 65 cases were found across 52 studies, dating back to 1958. This review represents the largest of sport-related structural brain injury associated with ACs to date. The clinical implications of our findings are discussed below.

**Presentation**

An overwhelming majority of cases (84%) were chronic, with a median time to presentation of nearly 4 weeks. Many cases had multiple presentations to clinic for lingering symptoms, and only on the second or third visit was imaging performed. However, this trend is subject to publication bias, as a delayed SDH due to sports injury is more novel than the more common acute SDH. In addition, it is evident from our review that sport-related structural brain injury associated with ACs is not only a problem in team sports. Sporting activities ranged from martial arts to winter sports (e.g., skiing, snowboarding), and these nonteam sports accounted for the majority of cases. Furthermore, it is important to realize that athletes engaged in noncontact sports have also been reported to have AC hemorrhage, necessitating a high level of attention to patients regardless of the contact level of sport played.

How these patients present is perhaps most interesting when differentiating structural brain injury from post-concussion syndrome (PCS). PCS is a well-studied entity across many populations after general trauma, military blast injury, and SRC. Tator et al. comprehensively described the phenotype of PCS across 138 athletes, where most patients were younger than 30 years with an average of 7.6 symptoms for a median of 6 months. As both populations have symptom duration of weeks to months, differentiating between a recovering PCS patient with no structural injury versus one with an AC associated SDH or hygroma becomes challenging.

Based on our review, a persistent headache was present in 98% of patients, nausea and/or vomiting in nearly half, and diplopia in 13%. This symptom constellation represents a clear picture of increased intracranial pres-
Sure (ICP). In the previously mentioned study of PCS, headache was also endorsed by 90% of patients, followed by memory disorders (58%) and concentration problems (57%); nausea was endorsed by 37% of athletes and vomiting in only 3%. The issue of when to order imaging after SRC is a controversial one, as imaging in the chronic phase of PCS can be low yield and costly. However, imaging was potentially lifesaving in the reviewed population of 65 patients. Thus, based on the present systematic review, we recommend imaging when persistent signs and symptoms of increased ICP are present. Based on a median time to presentation of 25 days, imaging can be pursued at any time, but there may be a higher yield weeks after injury. Differentiating between symptoms of increased ICP versus PCS is much easier written than accomplished, but a detailed temporal history, fundoscopic examination checking for papilledema, and neuropsychological testing may aid in this differentiation. Furthermore, 13 patients (21%) presented with an objective neurological deficit such as hemiparesis or facial weakness, which is never seen in PCS. Imaging should promptly be performed in all patients who demonstrate a fixed neurological deficit on clinical examination.

### Imaging and Treatment

Most structural injuries were seen as isodense or hypodense lesions, signifying the chronic nature of the subdural and/or intracystic hematoma. To our surprise, the AC location was difficult to standardize across many of the studies reviewed. The well-known Galassi classification quantifies ACs based on extent of middle fossa involvement and compression of adjacent neural structures. While some articles mentioned the Galassi type, most did not. Whereas some authors labeled an AC as middle fossa, others would call a similar lesion temporal. Thus, we were unable to delineate the AC location given the heterogeneous nomenclature used. We emphasize the need for future work of AC associated structural sport-related brain injury to standardize cyst location. Also, the size of the AC was not recorded in most studies, likely due to hemorrhage obscuring the ability to fully visualize the entire cyst. If possible, future studies should also include AC size details, as the relationship between AC size and hemorrhage is also unknown.

Open craniotomy was the most common mode of surgical treatment, and nearly half of all cases mentioned cyst fenestration. Fenestration to the basal cisterns was
low energy trauma. The incidental finding of an asymptomatic AC, which, when asymptomatic, have been viewed more as a relative than absolute contraindication. However, absolute contraindication is recommended in patients who are symptomatic, have brainstem compression, or harbor a syrinx. Although outside the scope of this systematic review, equally important are return to play issues in athletes with a ventriculoperitoneal shunt, epilepsy, or prior craniotomy. 

Future Directions

A more common problem than the case of a sport-related structural injury associated with ACs is the initial clinical discussion surrounding involvement in sports after the incidental finding of an asymptomatic AC. Unfortunately, minimal information is available in the literature to guide these decisions about sports participation with an AC. A postoperative outcome was mentioned in over half of the studies, often in a limited fashion, and all instances mentioned good or uneventful outcomes. Furthermore, only 3% of studies mentioned return to sport or activity. With the current estimate of asymptomatic ACs at 0.7%–1.7% of the population, it can be extrapolated that on an American football field comprising approximately 90 athletes from both teams, it would not be uncommon for at least 1 player to have an asymptomatic AC. The issue of involvement in sports with an AC, with or without structural injury, is an important one as surveillance imaging becomes increasingly used.

One report of a 32-year-old professional football player described an asymptomatic AC found on imaging for cervical radicular symptoms. The cyst was 7.5 cm in its greatest dimension and located in the left sylvian fissure. The player was cleared to play football and had no neurological sequelae, with normal findings on postseason MRI. Gamradt et al. postulated 2 questions: does the AC place the player at increased risk for a SDH or hygroma, and what are the sequelae of that structural injury? Anecdotally, the authors hypothesized the presence of an AC did confer a slightly higher risk of hemorrhage due to the cyst incompressibility and associated calvaria thinning, although the exact magnitude of the risk could not be quantified. Miele et al. echoed this sentiment, stating that although an AC may hold an increased risk of hemorrhage, it is not an absolute contraindication to participation in contact sports, and patients and family members should be counseled of these risks. Unfortunately, similar to other analyses, we cannot calculate incidence rates of hemorrhage, nor are we able to quantify risk.

To the second question, our systematic review yielded a somewhat incomplete answer. Of the 55% of studies that mentioned an outcome, all endorse an uneventful recovery. However, most studies devoted only one sentence to the postoperative course. Thus, based on the limited information provided, we can say that in those studies that reported outcomes, all patients recovered well at the gross neurological level. According to the current evidence base, we can say that in those studies that reported outcomes, all patients recovered well at the gross neurological level. Although outside the scope of this systematic review, equally important are return to play issues in athletes with a ventriculoperitoneal shunt, epilepsy, or prior craniotomy.
<table>
<thead>
<tr>
<th>Case No.</th>
<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Sport</th>
<th>Pathology</th>
<th>Midline Shift</th>
<th>Treatment</th>
<th>Outcome</th>
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<td>13, M</td>
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<td>Bur hole</td>
<td>NR</td>
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<td>11, M</td>
<td>Hockey</td>
<td>IPH</td>
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<td>No surgery</td>
<td>Good; no return to sport</td>
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<td>Raveenthiran &amp; Reshma, 2014</td>
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<td>11</td>
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<td>Hygroma</td>
<td>No</td>
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<td>Cress et al., 2013</td>
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<td>EDH</td>
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<td>Open craniotomy; cystoperitoneal shunt</td>
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<td>Yes</td>
<td>Bur hole</td>
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<td>NR</td>
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<tr>
<td>41</td>
<td>Mori et al., 2002</td>
<td>14, M</td>
<td>Physical/weight training</td>
<td>SDH</td>
<td>Yes</td>
<td>Bur hole</td>
<td>Good</td>
</tr>
<tr>
<td>42</td>
<td>Gelabert-González, 2002</td>
<td>13, M</td>
<td>Football</td>
<td>Hygroma</td>
<td>Yes</td>
<td>Open craniotomy</td>
<td>Good</td>
</tr>
</tbody>
</table>
view, biases of a retrospective review exist. First, publication bias may be present in 3 major forms: the predilection to publish on 1) good outcomes, 2) patients who underwent surgery, and 3) patients with delayed clinical findings. The push for publication may be a reason that all but 3 of the 52 studies reviewed described operative treatment, when in reality nonoperative treatment may occur more commonly than is presented. Furthermore, delayed (as opposed to acute) clinical findings are more likely to be published, as these tend to be more novel and reportable. Second, sport-specific data were sparse, and in some studies only a remote history of sport was mentioned, sometimes without a distinct impact or collision. Several studies reported unspecified athletic events, which could represent a wide range of sporting activities. Thus, we had to rely on the limited history provided. Third, operative details were not recorded uniformly across authors, and some studies lacked important detail in dictating operative treatment.

Conclusions

Sport-related structural brain injury associated with an AC is a rare occurrence. The current systematic review informs us that these injuries are often chronic, with median time to presentation of 25 days, and most commonly presenting with headache and nausea/vomiting. Injuries occur in both team/nonteam and contact/noncontact sports, and the majority are treated with open craniotomy or bur hole. Outcomes were good in all studies that mentioned one. That said, substantial information is needed to better treat and avoid these serious injuries. Cyst location was not standardized in most studies, nor was cyst size mentioned, making the relationship between cyst size and risk of rupture impossible to ascertain. Furthermore, significant gaps in postoperative outcomes and return to sport were found. According to the current evidence base, we do not feel that there is a clear contraindication to participate in sports in patients with an AC, although parents and children should
be counseled appropriately. Further studies are necessary to better evaluate AC characteristics that could pose a higher risk of adverse events after trauma.

**References**

28. Kertmen H, Gürer B, Yılmaz ER, Sekerci Z: Chronic subdu-

Disclosures
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Author Contributions
Conception and design: Zuckerman, Solomon, Sills, Bonfield. Acquisition of data: Prather, Yengo-Kahn. Analysis and interpretation of data: all authors. Drafting the article: Zuckerman. Critically revising the article: all authors. Reviewed submitted version of manuscript: Zuckerman, Solomon, Sills, Bonfield. Approved the final version of the manuscript on behalf of all authors: Zuckerman. Statistical analysis: Zuckerman.

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