Pericallosal Artery Aneurysm: systematic review
Aneurisma de Artéria Pericalosa: revisão sistemática

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ABSTRACT

Background: Pericallosal artery aneurysms, from A2 to A5 segments of anterior cerebral artery, are uncommon (literature states 2.3-9.2% of brain aneurysms). They have a high mortality rate and tend to rupture easily during exposure. This study was done with the intention of pointing out the described presentations of pericallosal artery aneurysms. Materials and Methods: A systematic review related to pericallosal artery aneurysm was performed in the PubMed, MedLine (Ebsco), LILACS and Scielo databases, using as keywords: “pericallosal artery”, “distal anterior cerebral artery” and “aneurysm”. Results: Pericallosal artery aneurysms are present mainly in patients over 50 years of age (2:1 female to male). In younger patients (less than 20 years old) the seizure is the most frequent symptom, while in adults (20-59 years old) it is the thunderclap headache. They commonly present with Hunt & Hess Grade (HH) II. However, 69% presented subarachnoid hemorrhage (SAH), presenting mainly with Fisher 4, which increases the chance of vasospasms. Conclusion: Thus, although the distal anterior cerebral artery (pericallosal) aneurysms display common symptoms, they usually have severe bleeding. Nonetheless, more studies are needed to analyze the epidemiological, demographic and clinical aspects of the pericallosal artery aneurysms.

Keywords: Pericallosal artery; Aneurysm; Distal anterior cerebral artery

RESUMO

Introdução: Aneurismas da artéria pericalosa, dos segmentos A2 a A5 da artéria cerebral anterior, são incomuns (a literatura aponta 2,3-9,2% dos aneurismas cerebrais). Possuem uma alta taxa de mortalidade e tendem a se romper facilmente durante a exposição. Este estudo foi feito com o intenção de apontar as apresentações descritas de aneurismas da artéria pericalosa. Materiais e Métodos: Foi realizada revisão sistemática relacionada aos aneurismas da artéria pericalosa no PubMed, MedLine (Ebsco), LILACS e Scielo, com as palavras-chave: “artéria pericalosa”, “artéria cerebral anterior distal” e “aneurisma”. Resultados: Aneurismas de artéria pericalosa apresentam-se principalmente em pacientes com mais de 50 anos de idade (2:1 feminino para masculino). Em pacientes mais jovens (menos de 20 anos), a convulsão é o sintoma mais frequente, enquanto em adultos (20 a 59 anos) é a cefaleia em trovoada. Comumente apresentam com Hunt & Hess (HH) II. No entanto, 69% apresentaram hemorragia subaracnoidea (HAS) apresentando-se principalmente com Fisher 4, que aumenta a chance de vasoespasmos. Conclusão: Assim, embora os aneurismas da artéria cerebral anterior distal apresentem sintomas comuns, geralmente apresentam sangramento grave. No entanto, mais estudos são necessários para analisar os aspectos epidemiológicos, demográficos e clínicos dos aneurismas da artéria pericalosa.

Palavras-chave: Artéria pericalosa; Aneurisma; Artéria cerebral anterior distal

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INTRODUCTION

The brain vascularization consists in two complementary circulations: anterior, coming from the bilateral internal carotid arteries (ICA); and posterior, coming from both vertebral arteries. Each ICA, into the cranial area, will ramify into three branches: the anterior cerebral artery (ACA), the middle cerebral artery (MCA) and the posterior communicating artery (PcomA)\(^3\). The ACA, goes anteriorly and medially, and will connect to its contralateral part, forming the anterior communicating artery (AcomA). This section, called A1 (or proximal ACA), will supply the anterior arm of the internal capsule and the head of the caudate. The distal part, located after the AcomA, is divided into another 4 segments along the corpus callosum, A2-A5, that united will form the pericallosal artery (PcalA)\(^3\). Some authors defend the beginning of the PcalA at the pericallosal-callosomarginal (PCCM) junction. However, Rhoton states that, given the variability of presence and location of origin of the callosomarginal artery, the pericallosal segment should be the distal portion of the ACA from the AcomA, which includes the A2-A5 segments\(^3\).

Even being rare, pericallosal aneurysms (literature states 2.3 to 9.2% of brain aneurysms)\(^4,5,6\) are still the second most common site of aneurysms on the distal anterior cerebral artery, mostly at the PCCM junction. Given that the anatomy of this artery follows the corpus callosum, it has a major angulation point around the genu\(^1\). As we move distally in the PcalA, the aneurysms will be more related to mycotic infections, traumatic and tumor embolus. Because of the limited subarachnoid space in this region, this type of aneurysm frequently presents interhemispheric subdural or intracerebral hematoma, and has higher incidence of bleeding than other locations. Its fragility and brain adherences increase these aneurysms premature rupture intraoperatively. Literature states that multiples aneurysms in this site is very common and unruptured ones have more chances of bleeding than in other locations\(^7\).

The purpose of this study is to gather and point out the described presentations of pericallosal artery aneurysms.

MATERIALS AND METHODS

Search Strategy
The systematic review was related to pericallosal artery aneurysm and its presentation and conducted from December 24 to December 25, 2018 using PubMed, MedLine (Ebsco), LILACS and Scielo databases. The keywords used were the association of “pericallosal artery” or “distal anterior cerebral artery” and “aneurysm” (and their Portuguese forms), and case reports and full-text were used as filters.

Study Selection
After the duplicates were removed, the articles were screened analyzing the following criteria: case reports; full article availability; occurred in human species; and in English, Portuguese or Spanish. Then, the authors modified the Jadad's Scale\(^8\) (Table 1) in order to define which articles were going to enter the study, and the studies received grades from 0 to 5. Only the reports with grade 5 entered the study. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) flow chart\(^9\) summing the articles selection is exposed below at Figure 1 and the included articles are demonstrated at Table 2.

The search strategy resulted in 118 articles on the PubMed database, 20 on Medline, 6 on Scielo and 6 on LILACS, making a total of 150 reports from which 39 were duplicates and, therefore, removed, remaining 111 cases. Thirty-five of those cases were removed because did not meet the full-text availability and language criteria. Of the remaining 76 articles, 33 were excluded because they did not fulfilled the analysis based on the Modified Jadad's Scale (Table 1) since they were 15 traumatic aneurysms, 1 mycotic aneurysm and 17 non-characterized (missing any of the proposed criteria at the scale). At last, 43 articles were included at the study, making a total of 59 cases.

Data Synthesis and Analysis
The cases were synthetized using a Microsoft Excel\(^*\) sheet discriminating the following aspects: age, gender, HH Grade\(^10\), Fisher Scale\(^11\), clinical presentation and outcomes. To analyze
the gender proportion, the authors divided the number of female patients over male ones.

Aiming to analyze the age stratification, the total range (∆T) was calculated by subtracting the lower limit of the upper; then, using one of Milone's criteria the number of intervals (k) was determined; at last, the range of classes was defined by dividing ∆T/k.

The formulas were:

\[ \Delta T = 81 - 0.13 = 80.87 \]

\[ k = \sqrt{n} = \sqrt{59} = 7.68 \approx 8 \text{ classes} \]

\[ h = \frac{\Delta T}{k} = 10.10 \approx 10 \text{ years} \]

| Question                                                                 | Yes | No |
|--------------------------------------------------------------------------|-----|----|
| The study demonstrated all relevant clinical data (presentation, age, gender)? |     |    |
| The study exposed or enable the authors to use the Hunt & Hess Scale?     |     |    |
| The study exposed or enable the authors To use the Fisher Scale?          |     |    |
| The study specified the aneurysm classification?                          |     |    |
| This classification was non-mycotic and non-traumatic?                    |     |    |

**Table 1.** Modified Jadad’s Scale

![Flow chart](image_url)
Table 2. Articles selected with number of cases analyzed and outcomes. (n= Number of cases analyzed).

| Paper                | n | Outcome | Paper                | n | Outcome |
|----------------------|---|---------|----------------------|---|---------|
| Alleyne Jr, 1997     | 1 | GR      | Koenigsberg, 2006    | 1 | GO      |
| Basuito, 2004        | 1 | GR      | Koyama, 2000         | 1 | D       |
| Castaño-Leon, 2014   | 1 | GR      | Lawton, 2006         | 1 | GR      |
| Choi, 2011           | 1 | GR      | Lee, 2017            | 1 | GR      |
| Correa, 2013         | 1 | GR      | Matsumoto, 2005      | 1 | GR      |
| Demartini Jr, 2016   | 1 | GR      | Mizunari, 2011       | 1 | GR      |
| Dhandapani, 2018     | 1 | GR      | Mori, 1995           | 1 | GR      |
| Dunn, 2011           | 1 | GR      | O'brien, 1997        | 1 | GR      |
| Enesi, 2013          | 1 | GR      | Park, 2008           | 1 | GO      |
| Erdogan, 2005        | 1 | GR      | Senbokuya, 2012      | 1 | GR      |
| Farias, 1997         | 1 | GR      | Sgulo, 2018          | 1 | GR      |
| Fathi, 2014          | 1 | GO      | Sousa, 2002          | 2 | GR/D    |
| Frank, 1990          | 1 | GR      | Tomar, 2017          | 1 | GR      |
| Gelfenbeyn, 2009     | 1 | GR      | Topsakal, 2003       | 1 | D*      |
| Hermann, 2014        | 2 | GR/D    | Ture, 2000           | 1 | GR      |
| Hsu, 2008            | 3 | GR (3)  | Vachhani, 2016       | 1 | GR      |
| Inci, 1998           | 4 | GR (3)/D| Wakabayashi, 2000    | 1 | GR      |
| Jagetia, 2007        | 1 | D       | Waldau, 2013         | 1 | GO      |
| Janjua, 2013         | 1 | GO      | Watanabe, 1991       | 3 | GR/D/D  |
| Kamiya, 1991         | 2 | GR/GR   | Xue, 2010            | 1 | GR      |
| Kaneko, 2001         | 1 | GR      | Yoneda, 2014         | 1 | GR      |
| Kim, 2007            | 7 | GR (ALL)|                     |   |         |

RESULTS

The cases were initially divided into 8 classes with a 10 years range, but, to better analyze and categorize the data, the authors united some of the classes reducing the number to four: < 20 years old (with 3 patients), 20-49 years old (15 patients), 50-59 years old (18 patients) and ≥ 60 years old (23 patients). The youngest patient described in our review was only 7 weeks and the eldest was 81 years old. Generally, most cases occurred in females over males (2.1:1) and 43 of those were ruptured aneurysm and the other 16 were unruptured. But as we divide per age range, the prevalence by gender, although, in all, the women were more affected, varies: in the group < 20 years old they were females; in the 20-49, this study found a 1.1:1 (female:male) ratio; in the 50-59, females also prevailed over male with a 1.25:1 ratio; and the last group showed that women were 4.75 times more affected than men. The proportion is displayed on Figure 2.

When analyzed by the HH Grade, the clinical grade for patients with aneurysm: 16 (27%) presented HH 0 (unruptured); 20%
(12) HH II; 17% (10) HH IV; 14% (8) HH III; 14% (8) HH V; and 8% (5) HH I. In patients under 20 years old (all female) two thirds present HH I and the rest HH 0. When analyzing those between 20 and 49 years old, 27% were unruptured (three men to one woman); 20% presented HH V (male over female 2:1); 20% HH II (two women to one man); 20% HH I (all women); and 13% HH IV (equally between genders). In those from 50 to 59 years old; 33% presented HH II (all male); 22% HH III (all female); 17% HH V (female over male 2:1); 17% weren't ruptured (two women to one man); and 11% presented with HH IV (both female). In the group of patients ≥ 60 years old, most of them had unruptured aneurysms (35%), being 8 women to 1 man, followed by HH IV (all female) with 26%; 17% presented HH III (equally between genders); 13% HH II (female over male 2:1); and only 9% HH V (all female). The frequencies are displayed on Figure 3.

As for the Fisher scale, which grades the SAH using the CT, only 41 cases were analyzed given that 16 weren't ruptured and 2 of the ruptured did not present SAH. Of the other 41 patients, 24 (59%) showed Fisher 4; 10 (24%) Fisher 3; 6 (15%) Fisher 2; and only one (2%) Fisher 1. However, one of the patients that presented Fisher 3 worsened to grade 4 while hospitalized (age range of 50 to 59 years). When analyzed by age, it was noted that one of the infants with less than 1 year presented Fisher 3, and the other, Fisher 4 (both females). Among the patients in the group 20-49, 50% of patients who bled presented Fisher 4 (male over female 1:5:1); 30% presented Fisher 3 (all females); 10% Fisher 2 (male); and 10% Fisher 1 (female). In the patient group 50-59, 60% presented Fisher 4 (female over male 1.25:1), 27% with Fisher 3 (female over male 3:1), 13% with Fisher 2 (all male) and none with Fisher 1. In elder patients (over 60 years), 64.3% presented Fisher 4 (female over male 2:1); 14.3% Fisher 3 (all female); 21.4% with Fisher 2 (all female); and none with grade 1. The proportion of cases by the Fisher scale is illustrated on Figure 4.

When analyzed by age range, in the group of patients < 20 years old, the most common symptom found was seizure (present in all of them), while in the group 20-49 years old, 47% of patients presented headache. The number rises to 61% (50-59 years old) and then falls to 35% (≥ 60 years old), when the presentation is more abrupt (such as SAH and ICH) and with focal deficits.

As for the outcomes, of the patients under 20: two were fully recovered and the other recovered with deficit. Of those from 20 to 49: 12 patients had great recovery (full), 2 recovered partially and one died. In those from 50 to 59, 13 were fully recovered, one partially recovered and 4 died (one from vasospasm, one from PTE, one from pulmonary infection and the other never recovered). In elderly patients (≥ 60), one partially recovered, 3 died (one from thrombophlebitis and the other two from complications of aneurysm rupture) and the other 19 were fully recovered.

The cases reported a variety of symptoms being the headache (HA) the most common one (29). Following the HA, the most common symptoms were focal deficits (6 patients); nuchal rigidity (6); seizures (6); and torpor (6). Other symptoms weren't as common as those above, such as hemiparesis (5); decerebrate rigidity (5); coma (5); emesis (5); syncope (5); and confusion (5). Less frequent ones included: four subdural hematomas, fever (3); hyperreflexia (3); memory deficits (3); nausea (3); infection associated (2); dizziness (2); gait disorder (1); hydrocephaly (1); ventriculomegaly (1); fatigue (1); vasospasm (1); vegetative disorders (1); personality alteration (1); and stupor (1). Only one patient presented totally asymptomatic. Nine patients also presented comorbidity. The most frequent were hypertension (5), followed by diabetes (2). Atrial fibrillation, drug abuse, migraine, cluster HA and polycystic kidney disease were also identified (all in one case each).
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HH 0 HH I HH II HH III HH IV HH V

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Figure 2. Frequency of patients age total and by gender.

Figure 3. Frequency of Hunt & Hess Grade cases by age range.

Figure 4. Frequency of Fisher Scale cases by age range.

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Through this systematic review it was possible to conclude that pericallosal artery aneurysms commonly present themselves as ruptured ones (73% of cases studied). The incidence on women were higher than in men, with a 2.1:1 ratio, but reached its peak of difference in the group of patients ≥ 60 years old, when women were 4.25 times more affected. There were two cases with age less than one year and the more frequent cases in patients above 50 years old (being 18 from 50 to 59 and 23 from 60 and over).

When analyzed the HH Grade, while generally most of the patients with ruptured presented with HH II grade (just as those from 50-59 group), the elderly patients (≥ 60 years old) presented mostly HH IV and the younger ones HH I. The reported symptoms varied in age range as well: while younger patients presented with seizures, generally, from 20 to 59 years old the most common symptom was the headache (reaching 61% of patients from group 50-59). As for the Fisher Scale, generally, and in all but one age range analyzed, the Fisher 4 was the most found (half of the younger patients presented 4 and the other half, 3). As for the outcome, only 13.5% of patients died; the rest 86.5% recovered well (8.5% partially and 78% fully).

Therefore, our study showed that pericallosal artery aneurysms, twice as often in women as in men, are present mainly in the patient over 50 years of age. Despite having a varied symptomatology, in younger patients the seizure is more frequent than the commonly associated symptom to aneurysms, the thunderclap headache. They commonly present with HH II. However, they also frequently bleed (69% presented SAH and 3% with SDH), presenting mainly with Fisher 4, which increases the chance of vasospasms. Furthermore, this review came to a lethality rate of 13.5%.

Thus, although the distal anterior cerebral artery aneurysms display common symptoms, they usually have severe bleeding. Nonetheless, more studies are needed to analyze the epidemiological, demographic and clinical aspects of the pericallosal artery aneurysms.
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