





Correlation of Clinical Presentations and Imaging Findings with Flow Dynamics in Carotid-Cavernous Fistula Patients at Dr. Hasan Sadikin Hospital, Bandung

Achmad Adam^{*}, Ingrid Ayke Widjaya¹

Department of Neurosurgery, Faculty of Medicine, Universitas Padjadjaran, Dr. Hasan Sadikin Hospital, Bandung, West Java, Indonesia

Abstract

BACKGROUND: Carotid-cavernous fistulas (CCFs) have variable clinical presentation, imaging, and angiographic findings.

AIM: This study aims to investigate the association of clinical presentations and radiological findings with flow dynamics from digital subtraction angiography (DSA) of CCFs patients. Knowing this association might be important to evaluate possible factors that have prognostic values in patients who are diagnosed with CCFs.

METHODS: CCF patients who underwent DSA at Dr. Hasan Sadikin general hospital from January 2017 to December 2019 were included in this study. Patient's characteristics, clinical presentations, and imaging results were retrieved from the patient's medical record and radiology database. Fractures, proptosis, extraocular muscle thickening, superior ophthalmic vein dilatation, cavernous hyperdense lesion, and infarct are expected to be identified from imaging results. DSA identified types of flow dynamic based on Barrow classification and venous drainage patterns. Numeric data were analyzed using Mann-Whitney U-test, while categorical data were analyzed with Fisher's exact test.

RESULTS: Twenty-eight patients were included in the study, with patients' mean age was 30.5-year-old (range: 14–61-year-old), consisting of 19 males (67.9%) and 9 females (32.1%). In approximately 75% of the cases, the cause of CCF was history of trauma. Patients with high-flow CCFs were associated with the findings of cavernous sinus hyperdense and proptosis than patients with low flow. Patients who are presented with more than 1-year long duration of symptoms were more likely to have more than 1 draining vein, compared to patients who are presented with <1-year long duration of symptoms.

CONCLUSIONS: History of trauma, longer duration of symptoms, and the presence of a hyperdense cavernous lesion on head CT scan results require further angiographic study before endovascular intervention.

Edited by: Branislav Filipović
Citation: Adam A, Widjaya IA. Correlation of Clinical Presentations and Imaging Findings with Flow Dynamics in Carotid-Cavernous Fistula Patients at Dr. Hasan Sadikin Hospital, Bandung. Open Access Maced J Med Sci. 2022 Mar 20; 10(B):1276-1279. <https://doi.org/10.3889/oamjms.2022.8659>
Keywords: Clinical and imaging findings; Flow dynamics; Endovascular; Carotid-cavernous fistula
***Correspondence:** Achmad Adam, Department of Neurosurgery, Faculty of Medicine, Universitas Padjadjaran – Dr. Hasan Sadikin Hospital, Bandung 40161, West Java, Indonesia. E-mail: achmad.adam@unpad.ac.id
Received: 17-Jan-2022
Revised: 25-Feb-2022
Accepted: 10-Mar-2022
Copyright: © 2022 Achmad Adam, Ingrid Ayke Widjaya
Funding: This research did not receive any financial support
Competing Interests: The authors have declared that no competing interests exist
Open Access: This is an open-access article distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 International License (CC BY-NC 4.0)

Introduction

Cavernous sinus (CS) is a venous plexus that receives drainage from the sphenoparietal sinus, superior ophthalmic vein, inferior ophthalmic vein, superior petrosal sinus, inferior petrosal sinus, and basilar venous plexus. Carotid-cavernous fistula (CCF) is characterized by abnormal communications between the CS and branches of either the internal carotid artery (ICA) or external carotid artery (ECA). Consequently, CCF patients may be presented with various symptoms, imaging, and angiographic findings [1], [2]. Based on its pathophysiology, CCF clinical symptoms can be classified into four groups, including orbital (chemosis, and proptosis), cavernous (ptosis, diplopia, ophthalmoplegia, and cranial nerves palsies), ocular (increased intraocular pressure, decreased vision, and eye pain), and cortical (headache, tinnitus, and ataxia) [2].

Based on its etiology, CCF may be spontaneous or traumatic. CCF might also be presented with high-flow and

low-flow dynamics. The barrow classification is arguably the most accepted system that has been used to classify CCFs based on their anatomical structures, in accordance with their arterial supply. Type A fistulas are defined as CCF that has direct communications between the cavernous segment of the internal carotid artery (ICA) and CS. The majority of cases with direct fistulas are usually accompanied by high-flow dynamic, usually acquired as a consequence of craniofacial fractures, aneurysmal rupture, or surgical intervention. Meanwhile, types B, C, and D are defined as CCF with indirect communications between CS and meningeal branches of the ICA or external carotid artery (ECA). Indirect fistulas are usually presented with unclear etiology. However, indirect fistulas appeared to be more prevalent in postmenopausal, diabetic, and hypertensive women. Indirect fistulas are usually featuring low-flow dynamics [2].

Based on their regions, patterns of venous drainage that is viewed on digital subtraction angiography (DSA) can be classified into four categories, including anterior (to superior and inferior ophthalmic vein),

posterior (to superior petrosal, inferior petrosal, and/or sphenoparietal sinus), inferior (to pterygoid plexus), and superior (to superior middle cerebral vein) [2].

Management of CCFs is based on their fistula types, degree of clinical symptoms, and risk of complications. Conservative treatment might be an option for patients with indirect fistulas, with an arguably low risk of complication. Meanwhile, for high-flow CCF, direct fistulas or patients with resistant clinical symptoms after conservative treatment of low-flow fistulas, endovascular, or surgical intervention should be considered [2]. This study aims to explore the association of clinical presentations and imaging findings with flow dynamics on DSA. Knowing this association would help clinicians to select cases that should be prioritized for DSA. Moreover, the results of this study will also suggest factors that have prognostic values in patients with CCFs.

Materials and Methods

This is a retrospective study of 28 cases of CCF at Dr. Hasan Sadikin Hospital during January 2017–December 2019. We recorded patient characteristics, clinical findings, imaging computed tomography (CT) scan study, and DSA findings. Numerical data were presented as mean \pm SD, while categorical data were presented as value. Categorical data were analyzed with Fisher's exact test. Numerical data were analyzed with independent t-test. All values are considered to be significant at $p < 0.05$.

Results

Characteristics of the subjects

Twenty-eight patients were included in the study, with patients' mean age was 30.5-year-old (range: 14–61-year-old), consisting of 19 males (67.9%) and 9 females (32.1%). In approximately 75% of the cases, the cause of CCF was a history of trauma. High-flow fistulas were identified in 21 (75.00%) patients. The difference in the fistula related to trauma between high-flow and low-flow CCFs (75.00% vs. 25.00%, $p < 0.0001$) was statistically significant. There was no difference in the duration of symptoms between patients with high-flow and low-flow CCFs (Table 1).

In our series, patients with high-flow CCFs were associated with eye protrusion ($p = 0.0002$) and chemosis ($p = 0.0007$) than patients with low-flow CCFs. The other symptoms were not distinguishing between high-flow and low-flow CCFs (Table 2).

Table 1: Association between patient characteristics with flow type

Variable	Flow type		p-value
	High flow (%) N = 21	Low flow (%) N = 7	
Age			0.9691
Mean \pm SD	29.50 \pm 14.14	29.75 \pm 16.13	
Median	26.00	27.50	
Range (Min-Max)	14.00–61.00	14.00–50.00	
Sex			0.6465
Male	15 (71.43)	4 (57.14)	
Female	6 (28.57)	3 (42.86)	
History of trauma			<0.0001**
(+)	20 (95.23)	1 (14.29)	
(-)	1 (4.76)	6 (85.71)	
Duration of symptom			0.6618
>1 year	11 (52.38)	5 (71.43)	
<1 year	10 (47.62)	2 (28.57)	

** $p < 0.0001$, with Fisher's Exact Test

Imaging findings

All patients underwent a head CT scan before DSA. Common findings on the head CT scan were CS hyperdense, proptosis, superior ophthalmic vein enlargement, and extraocular muscle thickening. Based on the angiography findings, the fistulas were classified according to their flow dynamic and drainage.

Table 2: Association between clinical findings with flow type

Variable	Flow type		p-value
	High flow N = 21 (%)	Low flow N = 7 (%)	
Eye protrusion			0.0002*
(+)	21 (100.00)	2 (28.57)	
(-)	0 (0.00)	5 (71.43)	
Redness			0.0764
(+)	15 (71.43)	2 (28.57)	
(-)	6 (28.57)	5 (71.43)	
Chemosis			0.0007**
(+)	16 (76.19)	0 (0.00)	
(-)	5 (23.81)	7 (100.00)	
Bruit			0.0825
(+)	19 (90.48)	4 (57.14)	
(-)	2 (9.52)	3 (42.86)	
Cranial nerve impairment			0.3642
(+)	9 (42.86)	1 (14.29)	
(-)	12 (57.14)	6 (85.71)	
Headache			0.6618
(+)	10 (47.62)	2 (28.57)	
(-)	11 (52.38)	5 (71.43)	
Blurred vision			>0.9999
(+)	11 (52.38)	4 (57.14)	
(-)	10 (47.62)	3 (42.86)	
Orbital pain			0.6219
(+)	4 (19.05)	2 (28.57)	
(-)	17 (80.95)	5 (71.43)	

* $p = 0.0002$, with Fisher's Exact Test, ** $p = 0.0007$, with Fisher's Exact Test

Early filling of the affected CS was identified in all patients. Patients with high-flow CCFs were associated with the findings of CS hyperdense ($p = 0.0432$) and proptosis ($p = 0.0002$) than patients with low flow (Table 3).

Table 3: Association between imaging findings on CT scan with flow type

Variable	Flow type		Nilai P
	High flow N = 21 (%)	Low flow N = 7 (%)	
Superior ophthalmic vein dilatation			0.3845
(+)	12 (57.14)	2 (28.57)	
(-)	9 (42.86)	5 (71.43)	
Cavernous hyperdense			0.0432
(+)	18 (85.71)	3 (42.86)	
(-)	3 (14.29)	4 (57.14)	
Proptosis			0.0002
(+)	21 (100.0)	2 (28.57)	
(-)	0 (0.0)	5 (71.43)	
Extraocular muscle thickening			0.6683
(+)	9 (42.86)	2 (28.57)	
(-)	12 (57.14)	5 (71.43)	

Duration of symptoms and number of draining vein(s)

The association of CCF's types and duration of symptoms with the number of draining vein(s) on angiographic studies was also analyzed (Table 4). Patients who are presented with more than 1-year long duration of symptoms were more likely to have more than 1 draining vein, with the mean number of draining veins 2.00 ± 0.852 (range 1–4) compared to patients who are presented with < 1-year long duration of symptoms, with the mean number of draining veins 1.06 ± 0.250 (1–2), $p = 0.0010$ (Table 4).

Table 4: Association between duration of clinical symptom with the amount of drainage pattern

Variable	Clinical symptom duration		p-value
	>1 year N = 16	<1 year N = 12	
Amount of drainage pattern			0.0010*
Mean \pm SD	2.00 \pm 0.85	1.06 \pm 0.25	
Median	2.00	1.00	
Range (Min-Max)	1.00–4.00	1.00–2.00	

* $p = 0.0010$, with independent t-test, $t=3.697$, $df=26$

Discussion

In line with the study by Ellis *et al.*, patients who were included in this study were predominantly young males (70%) [2]. CCF might be presented with a wide range of symptoms. Trauma is known as the most common cause of CCF. Cavernous ICA is strongly fixed by the dura when it enters and leaves the cavernous canal, immediately before the clinoids. Shear forces and traumatic shock waves tear the cavernous carotid artery, resulting in a high-flow fistula with massive arterIALIZATION of the CS [3], [4]. In line with this, the results of this study clearly showed that the presence of history of trauma is significantly associated with high-flow CCF type ($p < 0.0001$) (Table 1). Although redness, proptosis, and bruit are the three known classical symptoms of CCF, several symptoms can also confound the CCF diagnosis, including nerve palsies, headaches-associated conjunctivitis, and painful ophthalmoplegia. Here, we showed that history of trauma is significantly associated with the high-flow dynamic, thus should be considered when determining further treatment [5]. Furthermore, the results of this study confirmed the significant association of the presence of eye protrusion ($p = 0.0002$) and chemosis ($p = 0.0007$) with high-flow CCF (Table 2).

The presence of cavernous hyperdensity ($p = 0.0432$) and proptosis ($p = 0.0002$) on the affected side was acknowledged in the majority of cases included in this study and significantly associated with the high-flow dynamic type (Table 3). High cavernous pressure might be the cause of these imaging findings. This is in line with the study by Benson *et al.*, [6] which also identified CS asymmetry and or hyperdensity at

the affected site in the majority of patients. "Enlarged" CS was less commonly acknowledged among false-positive than true-positive cases. CCF often appears as an early enhancement of the CS on the side of the CCF, but not within the contralateral CS. Other imaging findings include dilatation of supraorbital vein, thickening of extraocular muscle, skull base fracture, and infarct [6].

DSA could identify the arterial supply and venous drainage patterns. The majority of cases included in this study were classified as high-flow CCF (Table 4). Anterior venous drainage pattern is the most common drainage pathway and might account for the predominant findings of orbital and ocular signs [1]. Analysis on correlation of duration of symptoms with amount of drainage pattern showed that the longer duration associates with higher number of drainage pattern. Jung *et al.* found that symptom duration before the procedure and number of venous drainage paths, later, correlated with recovery time after endovascular embolization. Early recovery (within 3 months) was more common in patients with a shorter symptom duration ($p = 0.043$) and fewer drainage paths ($p = 0.010$) [7].

In this study, we also noticed that patients with ocular symptoms are naturally brought to ophthalmologist before undergo CT scan, then DSA. Conservative treatment(s) include medication for ocular symptoms and compression of carotid artery and jugular vein ipsilateral to the CCF. These conservative treatments can be considered in patients with indirect CCFs (low flow; Type B and C) and patients with tolerable symptoms. Meanwhile, patients with direct CCFs (high flow; Type A) are less likely to achieve spontaneous resolution, thus have higher risks of developing progressive ocular symptoms, total loss of visual acuity, ischemic optic neuropathy, or even intracerebral and subarachnoid hemorrhage if left untreated [8]. The ideal management for these patients includes endovascular fistula-occlusion, such as detachable balloons, coils, embolic materials, and stents through a transarterial or transvenous route. In our study, history of trauma, fracture, and CS hyperdense viewed on head CT scan significantly correlated with the high-flow type. Therefore, further angiographic with urgent intervention is required if clinical or imaging findings suggested the signs of a high-flow CCF [2].

Conclusions

DSA remains the gold standard for diagnostic confirmation, management planning, and therapeutic intervention. All cases of CCFs should undergo an angiogram with selective cannulation of bilateral ECAs and ICA.

References

1. Timol N, Amod K, Harrichandparsad R, Duncan R, Reddy T. Imaging findings and outcomes in patients with carotid cavernous fistula at inkosi Albert Luthuli central hospital in Durban. *SA J Radiol.* 2018;22(1):1264. <https://doi.org/10.4102/sajr.v22i1.1264>
PMid:31754490
2. Ellis JA, Goldstein H, Connolly ES, Meyers PM. Carotid cavernous fistula. *J Neurosurg Focus.* 2012;32(5):9. <https://doi.org/10.3171/2012.2.FOCUS1223>
PMid:22537135
3. Korkmazer B, Kocak B, Tureci E, Islak C, Kocer N, Kizilkilic O. Endovascular treatment of carotid cavernous sinus fistula: A systematic review. *World J Radiol.* 2013;5(4):143-55. <https://doi.org/10.4329/wjr.v5.i4.143>
PMid:23671750
4. Zanaty M, Chalouhi N, Tjounmakaris SI, Hasan D, Rosenwasser RH, Jabbour P. Endovascular treatment of carotid-cavernous fistulas. *Neurosurg Clin N Am.* 2014;25(3):551-63. <https://doi.org/10.1016/j.nec.2014.04.011>
PMid:24994090
5. Castro LN, Colorado RA, Botelho AA, Freitag SK, Rabinov JD, Silverman SB. Carotid-cavernous fistula a rare but treatable cause of rapidly progressive vision loss. *Stroke.* 2016;47(8):207-9. <https://doi.org/10.1161/STROKEAHA.116.013428>
PMid:27406104
6. Benson JC, Rydberg C, DeLone D, Johnson MP, Geske J, Brinjikji W, *et al.* CT angiogram findings in carotid-cavernous fistulas: Stratification of imaging features to help radiologists avoid misdiagnosis. *Acta Radiol* 2020;61(7):945-52.
PMid:31698923
7. Jung K, Kwon BJ, Chu K, Noh Y, Lee ST, Cho YD, *et al.* Clinical and angiographic factors related to the prognosis of cavernous sinus dural arteriovenous fistula. *Neuroradiology.* 2011;53(12):983-92. <https://doi.org/10.1007/s00234-010-0805-3>
PMid:21161199
8. Yu SS, Lee SH, Shin HW, Cho PD. Traumatic carotid-cavernous sinus fistula in a patient with facial bone fractures. *Arch Plast Surg.* 2015;42(6):791-3. <https://doi.org/10.5999/aps.2015.42.6.791>
PMid:26618131