Clinical presentation, diagnosis and endovascular treatment of carotid-cavernous fistulas (CCFs): A single-centre experience

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Aims and objectives

A carotid-cavernous fistula (CCF) is a type of arteriovenous fistula, in which a pathologic communication exists between the carotid artery or its branches and the cavernous sinus. Such fistulas form spontaneously or due to secondary causes (e.g., trauma). CCFs occur quite rarely, but are important to note because in many cases they are associated with a marked ophthalmologic and neurologic morbidity and significant complications [1, 2].

Aim
The goal of this study was to gather and characterize data about patients with CCFs that were hospitalized and received endovascular treatment at Pauls Stradiņš Clinical University Hospital, Latvia.

Objectives

- Analyse the demographic data and clinical presentation of patients.
- Determine the techniques used for radiological diagnosis of CCFs and assess imaging findings.
- Describe and classify CCFs according to their features.
- Examine the treatment received by patients and its outcomes.
Methods and materials

This is a retrospective study. Patients with a diagnosis of a CCF hospitalized between 2002 and 2013 were selected from an electronic medical record (EMR) database. Relevant clinical data were obtained from patient medical records. Imaging data were reviewed by the use of a picture archiving and communication system (PACS). Descriptive statistical measures such as absolute and relative frequencies, means etc. were calculated from the collected data.
Results

Patient population
During the period of interest there were 13 patients that underwent treatment for a CCF. Patient age ranged from 21 to 89 years, mean age being 56 (±19) years. Five patients (38%) were male and eight (62%) were female. Patient distribution by age groups and sex can be seen in Fig. 1 on page 6.

Clinical presentation
Patients were evaluated by a consulting neurologist and/or neurosurgeon and an ophthalmologist. The clinical picture in most cases consisted of exophthalmus, conjunctival injection, ophthalmoplegia and visual disturbance with other symptoms being less common (see Fig. 2 on page 6). Signs and symptoms were ipsilateral for nine (69%) patients, contralateral in one (8%) patient and bilateral in two (15%) patients (Table 1 on page 7). A patient with bilateral fistulas exhibited only unilateral symptoms. The classical triad of symptoms (pulsating exophthalmus, conjunctival injection and bruit) was present in nine (69%) patients [2, 3].

Radiological diagnosis
The diagnosis of a CCF was based on a combination of the clinical picture and imaging findings. Depending on the clinical circumstances (i.e., patient's clinical status, time of symptom onset, speed of symptom progression and severity etc.) different imaging modalities were used. The employed techniques were computed tomography (CT), computed tomography angiography (CTA), magnetic resonance imaging (MRI), 3D time-of-flight magnetic resonance angiography (MRA) and digital subtraction angiography (DSA). Typical imaging findings on the non-invasive imaging modalities were exophthalmus with extraocular muscle enlargement and periorbital oedema, dilation of the superior ophthalmic vein and cavernous sinus, venous congestion and abnormal drainage, early contrast enhancement etc. (Fig. 3 on page 8, Fig. 4 on page 9, Fig. 5 on page 10, Fig. 6 on page 11, Fig. 7 on page 12, Fig. 8 on page 13). The diagnosis was always later confirmed by DSA.

CCF description and classification
To facilitate better planning of treatment (and due to other reasons) CCFs can be classified by features such as aetiology, haemodynamics (i.e., high or low flow) or anatomy [4]. It is common practice to use a classification scheme developed by Barrow et al. [5].
Of the diagnosed CCFs, nine (69%) were associated with previous trauma and four fistulas (31%) were deemed to be spontaneous in their origin. Anatomically speaking, eight fistulas (62%) were direct and five (38%) were indirect. Almost all CCFs (92%) were
unilateral, with the exception of one bilateral case. The findings are summarized in Table 2 on page 14 and Table 3 on page 15.

Treatment
The endovascular treatment of CCFs is tailored to each patient. The choice of treatment approach is governed by factors such as fistula type, embolic agent availability, operator experience, risk of potential complications etc. [4]. In the case of direct CCFs a transarterial approach was used, while for indirect fistulas a transvenous approach was preferred. The embolic agent in most cases was platinum coils. In the oldest case detachable balloons were used. Conversely in the most recent case a vascular stent-graft was deployed, which represents a fairly new approach to treatment of CCFs (Fig. 9 on page 16) [1, 6]. Unfortunately in five cases (38%) due to the extent or complexity of the underlying pathology it was necessary to sacrifice the internal carotid artery (ICA) in order to achieve technical success (Fig. 10 on page 17, Fig. 11 on page 18). Also as a result of fistula re-canalization and reoccurrence of symptoms, three patients (23%) required a repeat CCF closure procedure (Fig. 12 on page 19). All patients were discharged with an improvement in their clinical status, although in some cases with residual symptoms such as ophthalmoplegia, diplopia, vision loss etc.
**Fig. 1:** Patient distribution by age groups and sex.

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Fig. 2: Frequency of signs and symptoms.

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<table>
<thead>
<tr>
<th>Clinical manifestation</th>
<th>Fistula location</th>
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<tr>
<td>Unilateral</td>
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<tr>
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</tr>
<tr>
<td></td>
<td>Contralateral</td>
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<tr>
<td></td>
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</tr>
<tr>
<td></td>
<td>n/a</td>
</tr>
<tr>
<td></td>
<td>-</td>
</tr>
<tr>
<td>Bilateral</td>
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</tr>
<tr>
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<td>2</td>
</tr>
<tr>
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<td>0</td>
</tr>
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</table>

**Table 1:** Fistula location and clinical manifestations.
Fig. 3: CTA image showing indirect signs of a CCF. On the left side there is exophthalmus. The left superior ophthalmic vein is dilated and in its proximal part early contrast enhancement can be seen.

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Fig. 4: CTA volume rendering showing signs of venous hypertension due to a CCF.

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Fig. 5: A sequence of DSA images showing a direct, type A carotid-cavernous fistula due to trauma.

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Fig. 6: A sequence of DSA images after the closure of the previously demonstrated fistula with platinum coils.

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**Fig. 7:** A direct, type A carotid-cavernous fistula due to trauma. DSA image in the coronal plane.

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Fig. 8: DSA image after the closure of the previously demonstrated fistula with platinum coils.

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Table 2: Number of CCFs by their aetiology and anatomical type.

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Table 3: Patient demographic data and CCF classification.

<table>
<thead>
<tr>
<th>No.</th>
<th>Patient ID</th>
<th>Age</th>
<th>Sex</th>
<th>Fistula location</th>
<th>Aetiology</th>
<th>Anatomical type</th>
<th>Type according to Barrow et al.</th>
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<td>spontaneous</td>
<td>indirect (dural)</td>
<td>D</td>
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<td>indirect (dural)</td>
<td>B</td>
</tr>
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<td>indirect (dural)</td>
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<tr>
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<td>indirect (dural)</td>
<td>B</td>
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<tr>
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<td>89</td>
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<td>left</td>
<td>spontaneous</td>
<td>indirect (dural)</td>
<td>B</td>
</tr>
</tbody>
</table>
**Fig. 9:** DSA image before (A) and after (B) the closure of a CCF with a stent-graft in the internal carotid artery (ICA).

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Fig. 10: A sequence of DSA images of a CCF that required the sacrifice of the internal carotid artery (ICA).

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**Fig. 11:** DSA image after the occlusion of the internal carotid artery (ICA) with platinum coils.

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Fig. 12: The number of treatment sessions required per patient.

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Conclusion

The occurrence of only a small number of cases that were diagnosed and underwent endovascular treatment during the period of interest attests to the notion that CCFs are a rare condition.

Although no definite association between patient age and sex exists, it seems that there is a tendency for direct CCFs to occur more frequently in young males and for indirect fistulas to occur more frequently in older females. This observation has been expressed in other studies [1-3, 6-8].

Overall it can be concluded that endovascular therapy is an effective method of treatment for CCFs as it generally leads to good outcomes for patients.
References


