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The Direct Carotid Cavernous Fistula: a Clinical, Pathoanatomical, and Physical Study

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Summary

In order to further elucidate the pathogenesis of the direct carotid cavernous fistulas (dCCF) clinical, patho-anatomical, and physicomechanical studies were performed.

In 27 of 42 patients the dCCF were found to be localized in the segment C 4 (according to Teufel, 12), in 13 patients in segment C 2 and in only 2 patients in segment C 3. The patients with dCCF in segment C 4 were significantly younger than those with dCCF in the segments C 2 or C 3.

In none of the patients fractures of the bony walls of the cavernous part of the internal carotid artery (ICA) could be ascertained.

On human cadavers it was affirmed that the cavernous branches of the ICA arise nearly exclusively from the top of segment C 3 and from the lateral wall of segment C 2. The strength of the wall of the cavernous part of the ICA was shown to decline with age as revealed by means of a tensile machine. There were no significant differences between the four segments investigated.

As revealed by roentgenograms the distensibility of the ICA within its coverings was shown to be greatest in the segments C 2 and C 4 and lowest in segment C 3. A sudden increase of the intraluminal pressure ruptured the ICA exclusively in the segments C 2 and C 4. Histological preparations revealed that the trabeculae of the cavernous sinus insert tangentially into the adventitia of the ICA.

Taken together these findings strongly support the view that the dCCF are mainly due to a sudden increase of the intraluminal pressure of the ICA.

Keywords: Carotid cavernous fistula; pathogenesis.

Introduction

Direct carotid-cavernous fistulas (dCCF) are acquired arteriovenous shunts between the internal carotid artery and the cavernous sinus. They are thought to be due to a lesion of the wall of the cavernous part of the internal carotid artery (ICA) - or of the wall of one of its small branches.

It is generally accepted that the majority of the dCCF is caused by head trauma but only in a few cases a fracture of the base of the skull involving the cavernous part of carotid canal could be ascertained[8–10]. Similarly, in only a few cases the development of dCCF could be traced to tearing off of small carotid artery branches[2].

Since dCCF are agreed to prefer some well defined sites of the cavernous part of the ICA a common pathogenesis being related to anatomical conditions is strongly suggested. Since dCCF are very rare events individual predisposing factors are likely to be involved too.

In order to substantiate this view we present the data of 42 patients suffering from posttraumatic dCCF. In a second part we present the results of experiments performed on human cadavers concerning anatomical and physical conditions of the cavernous part of the ICA.

Material and Methods

1. Clinical Investigations

In 42 patients suffering from posttraumatic dCCF the localization of the fistula was determined by means of angiograms. The contrast medium was injected into the vertebral artery and forced to flow through the carotid-cavernous fistula by compressing the ipsilateral common carotid artery.

A fracture of the base of the skull was proven in 3 cases and suspected in 4 cases. In none of the cases included in this study, however, there was evidence of a fracture running through the cavernous sinus.

2. Laboratory Investigations

In 145 post mortem preparations of the internal carotid artery the anatomical (2.1.) and mechanical (2.2. and 2.3.) properties of the cavernous part of the internal carotid artery were investigated.

2.1. In order to study the cavernous branches of the internal carotid artery this artery was dissected within its bony coverings out of the base of the skull. The cervical stump of the artery was connected to a pvc-catheter and its supraclinoid end was ligated. Contrast medium (Urovision®, Schering) was pushed into the artery and a roentgenogram was performed.

Thereafter casts of the artery and its branches were produced by means of a red resin which was injected through the same catheter and which hardened within one hour (Technovit, Kulzer, Germany). The organic tissue was macerated by means of potassium hydroxide.

2.2. In order to discover the conditions responsible for rupture of the cavernous part of the ICA the mechanical properties of the vessel wall were determined by means of a tensile testing machine usually applied to industrial materials (Universal-Zugprüfmaschine RKM 100, Röll and Korthaus, Germany).

To avoid the effects of autolytic disintegration of the texture of the arterial wall the arteries were investigated not later than 50 hours after death[1]. The artery was dissected into four segments according to the proposal of Teufel[12] (Fig. 2). The segments were cut into longitudinally and into transversely aligned strips.

The strips were clamped into the clamping device of the tensile testing machine and were subjected to strain in the direction of the tensile force in uniform speed. The force-elongation curve was plotted until the force at break or the elongation at break, respectively, was reached. From these curves the break energy (Newton x meter) was determined.

2.3.1. Finally we tested why the cavernous part of the ICA ruptured preferentially in segments 2 and 4. In 21 experiments a PVC-catheter was inserted into the proximal stump of the artery and the artery was filled with contrast medium. Thereafter, the supraclinoid stump was ligated tightly. Then the intra-arterial pressure was increased slowly by means of a syringe. During the phase of pressure increase a series of roentgenograms were taken in order to determine the sites of maximal arterial dilatation.

2.3.2 In 22 experiments the arteries were prepared as described in section 2.3.1.. The intramural pressure was increased so steeply that the artery wall was overexpanded and ruptured. The localisation of the rupture was noted.

2.4. In 88 internal carotid arteries the distribution of arteriosclerotic calcifications was determined by means of native roentgenograms.

2.5. In order to visualize the region where the trabeculae of the cavernous sinus insert into the adventitia of the ICA several segments of the cavernous part of the ICA left within their bony coverings were embedded in paraplast. The histological sections were stained with the trichrome procedure.

For statistical evaluation student's t-test was used.

Results

1. Clinical Investigations

As revealed by angiograms posttraumatic fistulas developed most frequently in segment C4, their frequency was half as high in segment C2 and fistulas localized in segment C3 were detected in only 2 of the 42 patients. The mean age of the 27 patients with dCCF localized in segment C4 was 31 yrs (sd = 18) and that of 13 patients with dCCF in segment C2 was 50 yrs

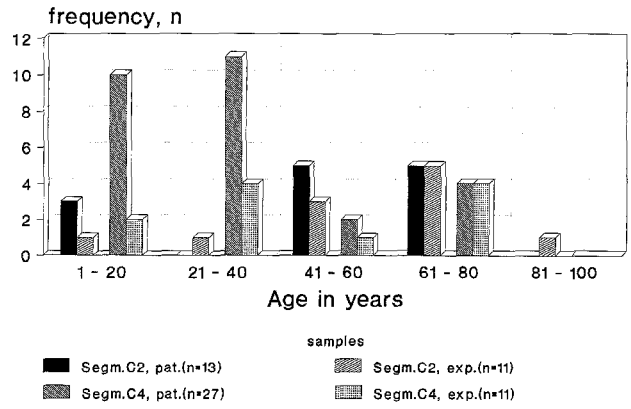

Fig. 1. The age distribution of dCCF and experimental ruptures in segments C2 and C4, respectively. The schematic segmentation of the artery is modified according to Teufel[12]

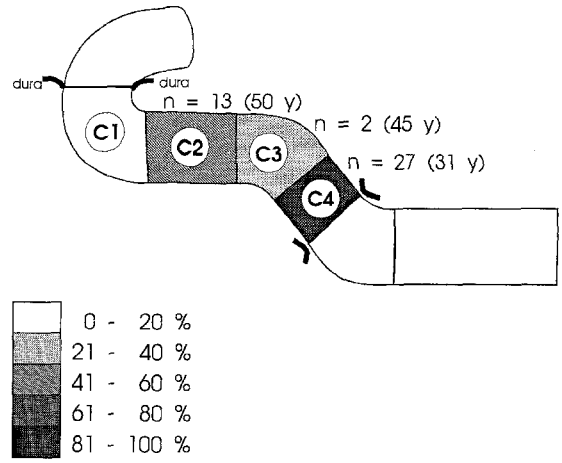

Fig. 2. The distribution of dCCF on the 4 segments of the cavernous part of the internal carotid artery

(sd = 20). The difference is highly significant ($p < 0{,}001$, Fig. 1 and Fig. 2).

2. Laboratory Investigations

2.1. As shown in Fig. 3 the distribution of the branches of the ICA revealed by roentgenograms equalled that observed in the casts. In more than 90% of the ICA investigated one or more branches arose from the dorsal aspect of segment C3 and in about 80% branches arose from the lateral wall of segment C2. In 2 of the very young children we detected small branches arising from the medial wall of segment C4.

2.2. The evaluation of the strain test revealed that the break energy required for rupturing of the tangential strips declined in all segments with age. This ten-

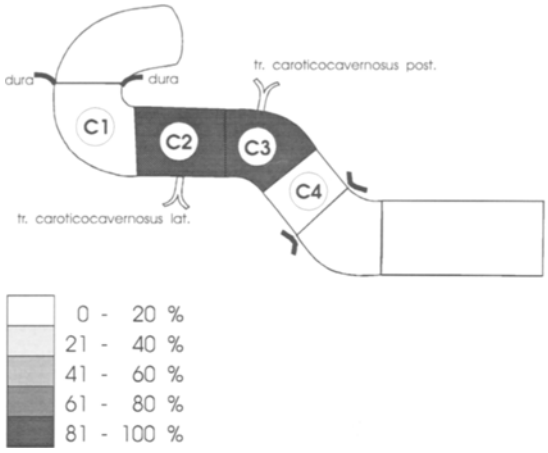

Fig. 3. The sites of origin of the small branches of the cavernous part of the internal carotid artery as revealed by roentgenograms and casts

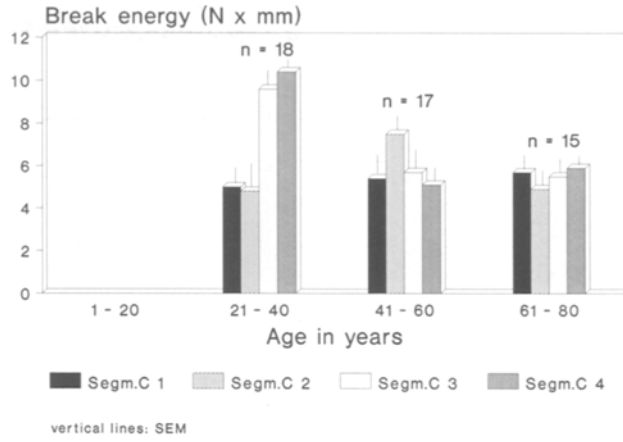

Fig. 5. The age distribution of the energy required to break the 4 segments of the cavernous part of the internal carotid artery when expanded longitudinally

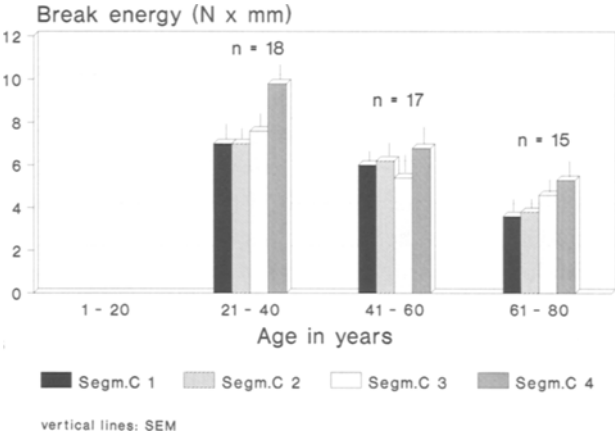

Fig. 4. The age distribution of the energy required to break the 4 segments of the cavernous part of the internal carotid artery when expanded transversely

Fig. 6. Transverse histological section of the cavernous part of the internal carotid artery (ICA) within its coverings (trichrome stain, X 30); arrow: trabeculum inserting into the adventitia of the ICA

dency was most clearcut for segment C 4 where the mean value of the 21 – 40-year-old group exceeded that of the 61 – 80-year-old group significantly ($p < 0{,}01$, Fig. 4).

The strength of the longitudinal strips appeared not to depend on age except for segments C 3 and C 4 of the specimens of the 21 – 40-year-old which broke at a significantly higher energy level than the other segments of the same and of all other age groups (Fig. 5).

2.3.1. Inflation of the ICA by injection of contrast medium revealed consistent differences in the regional distensibility of the vessel wall. In segment C 2 as well as in segment C 4 the maximal increase of the vessels diameter was about 25% of the initial value. This difference is highly significant ($p < 0{,}001$). Segment C 3, in contrast, retained its initial shape almost completely.

2.3.2. The ruptures resulting from a sufficiently steep increase of the intraluminal pressure were localized exclusively in the lateral wall of segment C 2 and C 4, respectively. The mean age of the 11 donors of those arteries which ruptured in segment C 4 was 40 yrs (sd = 28) and that of the 11 donors of the arteries which ruptured in segment C 2 was 61 yrs (sd = 26). This difference is not significant ($p < 0{,}09$, Fig. 1).

2.4. In about 94% of the 88 arteries investigated arteriosclerotic calcifications were localized in the outer curvature of segment C 1 and in about 80% segment C 3 was involved. In about 70% the calcifications extended into the segments C 2 and C 4 which, however, were involved in no case separately.

2.5. The histological investigations revealed that the trabeculae of the cavernous sinus inserted tangentially into the outermost layer of the adventitia of the internal carotid artery. In no case did they reach the muscular layer of the artery (Fig. 6).

Discussion

Our clinical data confirm that dCCF are more frequent in the young than in the elderly probably reflecting the larger risk of the former to experience head trauma. Further, it turned out that dCCF of the younger patients are preferentially localized in segment C 4 of the cavernous part of the ICA and those of the older in segment C 2. Segment C 3 appeared to be only rarely involved. To our knowledge there are no comparable data in the literature (see[4]).

Concerning the pathogenesis of the dCCF our findings do not support the view that fractures of the base of the skull represent the main pathomechanism[3, 6, 7, 11]. None of our 42 patients had an unequivocal history of skull fractures. Friedmann *et al.*[5] stressed that dCCF occurred in less than 1% of injuries of the skull and that their incidence did not parallel with that of head trauma.

Another pathomechanism suggested as cause of dCCF is tearing off of the small branches of the ICA[2]. We could affirm that the small branches of the cavernous part of the ICA regularly arise from the lateral wall of segment C 2 (tr. caroticocavernosus lat.) and from the superior aspect of the posterior curvature of segment C 3 (tr. caroticocavernosus post.). Since segment C 4 of adults was always free of branches this pathomechanism may apply only to those dCCF which are localized in segment C 2 and C 3. The tearing off-hypothesis, however, does not explain why the segment C 3 is so rarely involved in the genesis of dCCF and why the majority of dCCF is localized in segment C 4.

Thus, fractures of the base of the skull and tearing off of small branches of the ICA may be responsible for the pathogenesis of dCCF but only in some few cases.

Further, our findings do not support the view that the trabeculae which are spanned out between the outer surface of the ICA and the outer wall of the cavernous sinus may tear the wall of the ICA when stretched by traumatic distorsion of the base of the skull. The trabeculae are shown to insert tangentially into the adventitia of the ICA not reaching the muscular layer (Fig. 6). Thus it appears unlikely that tugging of the trabeculae may destroy the wall of the ICA.

Alternatively, we showed that a sudden increase of the intraluminal pressure of the ICA with the distal intracranial end of the artery being tightly ligated forced the vessel wall to rupture in segments C 2 and C 4; that is exactly at those sites known to be prefered by dCCF in vivo (Fig. 2). Moreover, it appeared that also under these experimental conditions the ruptures of the vessel wall in segment C 4 predominated in the younger age group. This difference, admittedly, was not statistically significant.

These results suggest that the majority of dCCF is due to a sudden increase of the intraluminal pressure of the ICA rather than to trauma to the vessel wall for instance by bony spikes or by tearing off of the small branches or tugging of the trabeculae.

The question remains why dCCF develop in segment C 2 and C 4, and almost never in segment C 3.

Concerning the resistance of the vessel wall to tearing forces a tendency to decline with age could be recognized for all segments (Figs. 4 and 5) and the longitudinal strips of the segments C 3 and C 4 showed a singificantly higher strength in the 21–40 year-old group than in all other age groups (Fig. 5). Apparently this finding can hardly explain the different behaviour of the segments C 3 and C 4.

If a sudden increase of the intraluminal pressure is the force responsible for the development of dCCF then it appeared justified to assume that it is the degree of distensibility of the ICA which defines the site of the rupture.

In cadavers we showed by means of roentgenograms that an increase of the intraluminal pressure distended segment C 2 and segment C 4 considerably whereas the shape of segment C 3 remained nearly unaltered.

This affirms the assumption that dCCF develop from direct rupture of the vessel wall and that the ruptures are mainly due to a critical increase of the tension of the vessel wall induced by an increase of the intraluminal pressure.

Critical increases of the intracarotid pressure may be induced by an intense axial acceleration of the body or by sudden compression of the carotid arteries as for instance by extreme and sudden extending or bending of the neck. The pressure wave runs cephalad and is likely to break at first on the wall of segment C 4. This may explain why the majority of the traumatic dCCF – especially of the young – are localized in this segment. It should be remembered that in some very

young children we have observed thin branches arising from segment C 4. These possibly represent remnants of the primitive trigeminal artery. Although these branches become atretic in later life they may leave behind some weak point in the vessel wall forming a locus of reduced mechanical resistance.

Finally we have to explain why the prefered localization of dCCF appears to shift with increasing age from segment C 4 to segment C 2 (Figs. 1 and 2). Since arteriosclerotic lesions are well known to develop around the orifices of arterial branches the wall of segment C 2 with its branches is more prone to be weakened by arteriosclerotic plaques than that of segment C 4 which is shown to be free of branches. Thus in the elderly with advanced arteriosclerosis segment C 2 may be ruptured by increases of the intraluminal pressure which do not suffice to endanger segment C 4.

In conclusion, our data demonstrate that there is a considerable congruency in localization and age of development between traumatic dCCF and experimental ruptures of the wall of the cavernous part of the ICA induced by a sudden increase of the intraluminal pressure. It further turned out that it is not the texture of the vessel wall but the width of the perivascular manchette built by the cavernous sinus which determines the site of rupturing of the vessel wall. Thus, in most cases traumatic dCCF appear to be due to a sudden critical increase of the intraluminal pressure of the ICA rather than to fractures of the base of the skull or to tearing off of the small branches of the ICA. With increasing age arteriosclerotic lesions of the wall of the ICA are thought to play an additional and localizing role in the pathogenesis of dCCF.

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