

Segmental agenesis of the internal carotid artery: angiographic aspects with embryological discussion

P Lasjaunias and A Santoyo-Vazquez

Service de Radiologie Adulte, Hôpital de Bicêtre, Université Paris XI, 78, rue du Général Leclerc, F-94270 Le Kremlin-Bicêtre, France

Summary. The embryology of the internal carotid artery (ICA) shows that this vessel comprises from origin to termination six segments, i.e. cervical, petrous, vertical cavernous, horizontal cavernous, clinoid and cisternal segments. Each of these segments displays a specific course and limits, defined by the origin of the following embryonic arteries: ventral pharyngeal hyoid, mandibular, primitive maxillary, trigeminal, dorsal ophthalmic and ventral ophthalmic. Each segment is independent and may show agenesis. In such cases the internal carotid blood flow (hemispheric arterial supply) is rerouted to afford usual ICA supply distal to the agenic segment.

All congenital anomalies of the ICA can be described and understood on the basis of embryological data. The “aberrant internal carotid” can therefore be identified as a normal vessel. Differentiation can be made between congenital versus acquired absence of the ICA. This type of analysis should allow the clinician to recognize what are normal, albeit rare variations, rather than to mistake them for an abnormal condition requiring treatment.

Agénésie segmentaire de la carotide interne. Aspects angiographiques, discussion embryologique

Résumé. L'étude de l'embryologie de la carotide interne montre qu'on peut la scinder en six segments consécutifs: cervical, pétreux, caverneux vertical, caverneux horizontal, clinôidien, cisternal. Chacun d'eux a un trajet et des limites fixes, marqués par l'origine de vaisseaux embryonnaires particuliers: pharyngée ventrale, hyostapédienne, mandibulaire, maxillaire primitive, trijémée, ophthalmique dorsale, ophthalmique ventrale.

Chaque segment est indépendant et peut être agénétique; le flux carotidien est alors “déviié” par un autre chemin pour ce seul segment, après quoi, il retrouve la carotide interne stricto sensu dans son trajet habituel. Différentes agénésies segmentaires sont ainsi présentées: cervicale, pétreuse, cavernueuse. Les “déviation” artérielles (cavité tympanique, fosse postérieure, côté opposé) et les vaisseaux embryonnaires objectifs (hyoïdienne, trijémée, maxillaire primitive) permettent de caractériser de façon anatomiquement précise le défaut artériel segmentaire et la solution hémodynamique qu'il a entraîné. On est dès lors capable de faire la différence entre une absence congénitale et acquise de la carotide interne, et celle qui sépare une disposition rare mais normale, prévisible et connue d'un aspect ignoré et considéré parfois à tort comme pathologique.

Key words: Embryology – Internal carotid artery – Agenesis – External carotid artery – Trigeminal artery

The aim of this paper, based on the study of a recent clinical case, is to achieve a brief didactic synopsis of agenesis of the internal carotid artery (ICA) and to draw attention to the practical importance of identification of the anomaly.

Agenesis of the internal carotid artery has been recognized for quite some time. In a study of the literature on this subject until 1980, Handa et al. [7] reported 45 published cases of unilateral absence of the ICA and 11 cases of bilateral agenesis. In most cases the circle of Willis, which is unaffected by this embryonic anomaly, allows revascularization of the homolateral cerebral hemisphere. Conversely, analysis of the literature seems to indi-

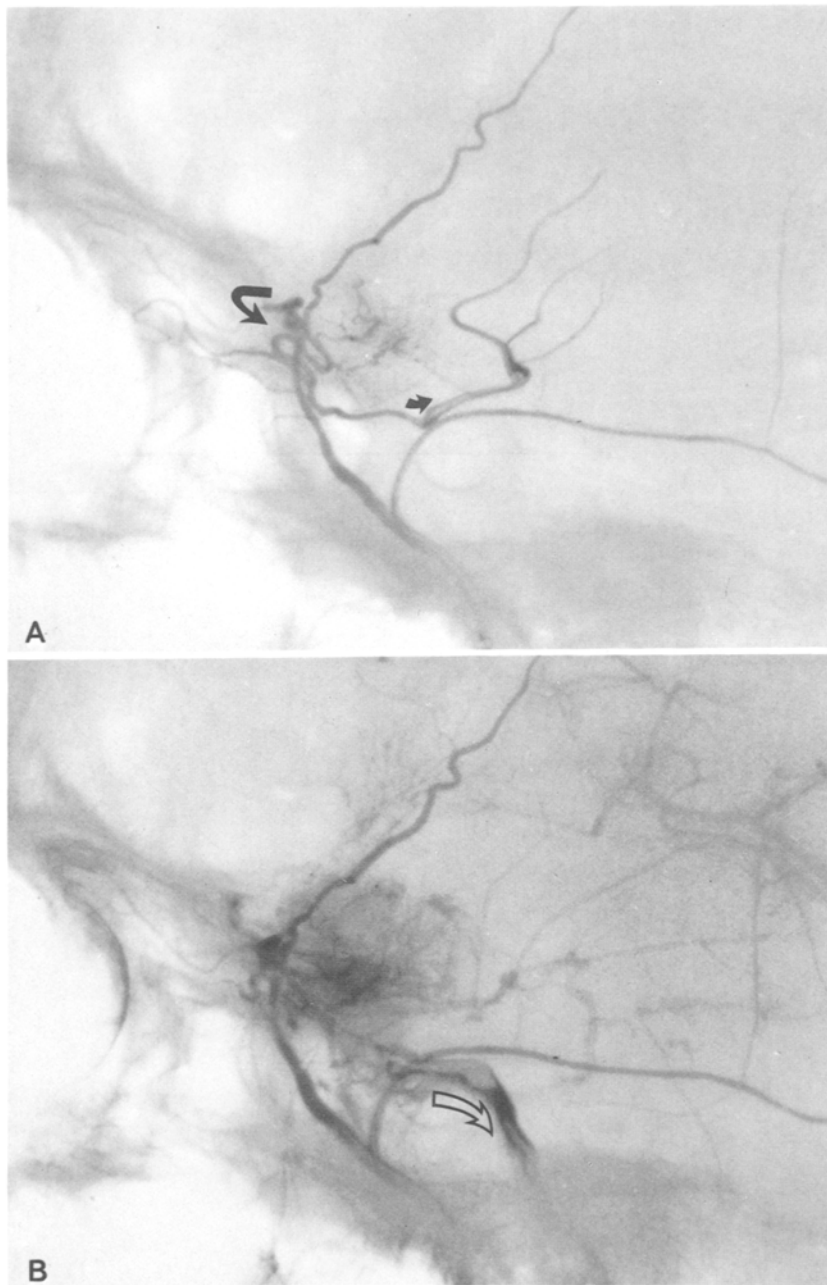


Fig. 1A, B

Selective opacification of the middle meningeal artery seen on lateral X-rays: **A** early filling; **B** late filling. At an early stage of opacification, the anastomosis between the frontal branch of the middle meningeal artery and the ophthalmic artery (*broken arrow*) allows counter current opacification of the supraclinoid carotid flexure (*small arrow*). The late films show the venous drainage of the tumor via the cavernous sinus (*curved outlined arrow*)

Injection sélective de l'artère méningée moyenne en incidence de profil: **A** temps précoce, **B** temps tardif. Sur le temps précoce, l'anastomose entre la branche frontale de l'artère méningée moyenne et l'artère ophtalmique (*flèche brisée*) réinjecte à contre courant le siphon carotidien supra-clinoïdien (*petite flèche*). Sur les temps tardifs, le drainage veineux de la tumeur s'effectue dans le sinus caverneux (*flèche courbe creuse*)

cate that segmental agenesis of the ICA occurs or is identified less frequently.

We report herein a case of segmental agenesis of the ICA, discovered fortuitously during roentgenological investigation of an intracranial tumor. To our knowledge, this case is the first published report of agenesis of the cervical segment of the ICA with intracavernous revascularization via the trigeminal artery. This paper presents the practical conditions having led to diagnosis, followed by a discussion of the clinical problem and finally a syn-

opsis of the different types of agenesis published in the literature.

Case study

Mrs H., 35 years old, was admitted to the department of Pr Lapresle, Hôpital de Bicêtre, in November 1982 because of generalized epilepsy of four months' duration. Clinical history revealed a cutaneous angioma of the mandibular region

treated by irradiation at the age of four months and thyroidectomy at the age of 26 years for asymptomatic goiter. Clinical examination showed no abnormalities and the results of laboratory investigation and standard skull X-rays were considered normal at that time. Electroencephalography demonstrated multiple spike waves in the left temporal region. Radionuclide scintiscanning showed pronounced uptake of tracer within the first hour in the left temporofrontal region. Computed tomography (Fig. 7), before and after contrast enhancement, revealed uptake of contrast material in the left anterior clinoid region extending towards the edge of the sphenoid bone. CT cuts made higher up demonstrated a significant parenchymal hypodensity of the frontal lobe. The working diagnosis was meningioma or brain metastasis. Selective carotid angiography was done as a diagnostic and pretherapeutic procedure. As a result of this investigation a diagnosis of meningioma was considered highly probable and the patient underwent surgery. Histological examination of the surgically removed tumor confirmed that it was meningioma.

Routine angiography in our service was performed under general anesthesia. Catheterization was via the femoral route with hyperselective injection of contrast medium. Catheterization of the left common carotid artery was achieved rather easily despite its small caliber noted from the onset. Diagnosis required that both the internal carotid and middle meningeal vascularization be opacified, and thus the middle meningeal artery was injected first (Fig. 1). The diagnosis of meningioma was confirmed by the demonstration of a parenchymal tumor in the clinoid region. Identification of the internal carotid artery via catheterization of the vessel originating from the left part of the aortic arch was unsuccessful. Furthermore, injection of contrast medium a few centimeters above the aortic origin of the vessel considered to be the common carotid artery did not demonstrate any cerebral branches (Fig. 2). In an attempt to identify the source of arterial vascularization of the left hemisphere successive catheterization of the contralateral ICA and left vertebral artery (the dominant side) was performed. Injection of the right ICA led to opacification of the anterior cerebral territory on the opposite side, opacification being via an azygos artery (Fig. 3) Injection of contrast material into the left vertebral artery demonstrated the presence of a trigeminal artery (Fig. 4). The latter was seen to arise from the distal part of the basilar artery and to run, as usually described, towards the cavernous sinus on the left side where it joined the ascending portion of the cavernous



Fig. 2
Opacification of the common carotid artery showing the infero-lateral trunk and intracavernous carotid flexure (*asterisk*). Note the pedicle supplying the tumor (*arrowhead*) and the absence of cerebral branches of the injected artery

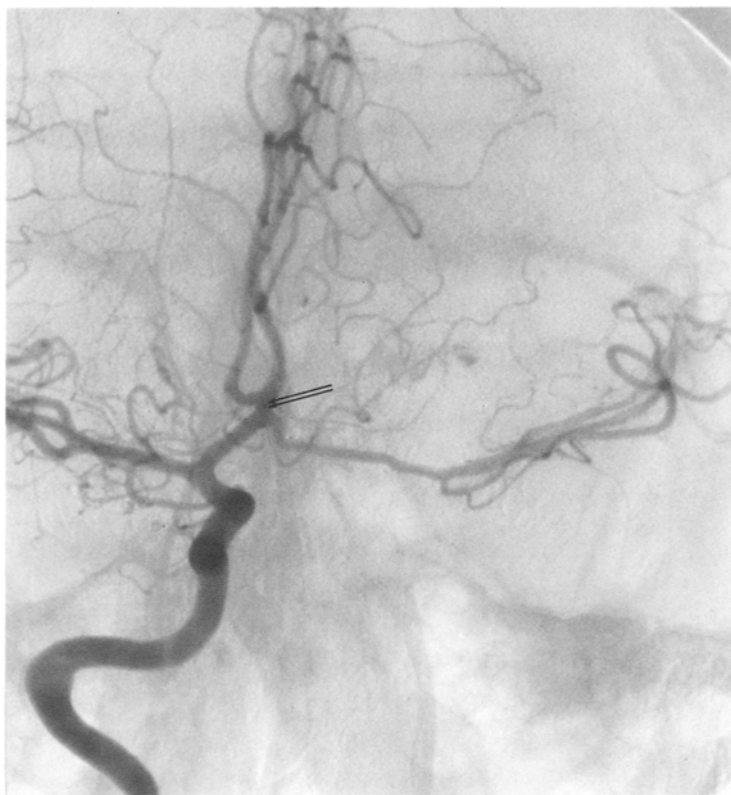
L'injection carotidienne primitive visualise le tronc inféro-latéral et un siphon carotidien intracaverneux (*astérisque*) ainsi qu'un pédicule nourricier de la tumeur (*pointe de flèche*). Aucune artère à destinée cérébrale n'est visible

flexure of the ICA. The distal part of the carotid flexure, its emergence from the cavernous sinus and the termination of the artery were those normally seen on angiography of the ICA.

The results of the angiography confirmed the diagnosis of meningioma of the anterior clinoid process and accompanying segmental agenesis of the ICA with intracavernous revascularization via a trigeminal artery.

Discussion

Retrospective analysis of the patient's standard skull X-rays showed the presence of hyperostosis

**Fig. 3**

Anteroposterior arteriogram via catheterization of the contralateral internal carotid artery. Note the opacification of an azygos artery (*twin arrows*)

L'injection carotidienne interne contro-latérale de face opacifie une artère azygos (*double flèche*)

of the anterior clinoid process. In a study of 42 meningiomas of the sphenoid ridge reported by Pompili et al. [14] hyperostosis was the most constant sign seen on standard skull roentgenograms. Hyperostosis reflects the bony invasion by the tumor and may be overlooked when the meningioma lies at the medial third of the sphenoid ridge. In the case of our patient, the history of radiotherapy may have been suggestive of post-irradiation meningioma, as recently discussed by Soffer et al. [15]. Likewise, owing to the absence of histological examination of the surgically removed goiter, it was conceivable that the patient's brain lesion was of metastatic origin, especially since the white matter adjacent to the lesion showed the presence of a hypodensity.

Regarding the question as to the acquired or congenital nature of the missing carotid artery, the history of radiotherapy suggested that occlusion may have occurred. However, CT scanning of the petrous bone demonstrated the absence of the carotid canal (Fig. 5) and thus confirmed that the nonvisualization of the ICA was of congenital origin. Indeed, the orifice of the canal at the base of the skull is absent when the artery normally contained within it is missing.

Embryological study of the ICA demonstrates

that this vessel results from the persistence of several embryologically different segments of fixed course and limits. Knowledge of these classical embryological features allows one to identify the different types of agenesis and the corresponding hemodynamic response leading to revascularization of the homologous cerebral hemisphere. To our knowledge, there have been no reports in the neuroradiological or anatomical literature referring to a hemodynamic response to agenesis of the ICA via the persistence or secondary development of the primitive stapedia, mandibular or trigeminal arteries. Thus, the case presented herein would be the first report of a trigeminal artery ensuring vascularization of a territory of the ICA owing to agenesis of the cervical and petrous segments of this vessel.

An intratympanic course of the ICA was often considered in the past as an unusual loop of the artery or even an aneurysm and was thus often ligated for this reason. One of us (PL) published a report in 1978 showing that the anomalies associated with an intratympanic course of the ICA, in addition to certain anatomical patterns in other Vertebrates, should lead to consider that this aberrant course of the ICA reflects the existence of segmental agenesis. This variety of agenesis can



Fig. 4

Lateral arteriogram with catheterization of the vertebral artery. A trigeminal artery can be seen to fill the intracavernous carotid flexure at the point of origin of the inferolateral trunk (*asterisk*) (Fig. 2). A branch supplying the tumor is also visible (*arrow-head*)

Injection de l'artère vertébrale en incidence de profil; une artère trijémminée opacifie le siphon carotidien intra-caverneux au point d'origine du tronc inféro-latéral vu sur la fig. 2 (astérisque). De même la branche à destinée tumorale est opacifiée (pointe de flèche).

be described as follows: the common carotid artery gives off the usual branches of the external carotid artery; the ascending pharyngeal artery, via its inferior tympanic branch, penetrates through Jacobson's canal (tympanic canal) into the tympanic cavity. At this point, the vessel which is normally anastomosed to the caroticotympanic artery (*rami carotico-tympanici arteriae carotis internae*) enters the intrapetrous carotid canal to supply arterial blood to the homolateral hemisphere. These observations help to understand some of the anomalies associated with this uncommon anatomical arrangement: the absence of an extracranial orifice of the intrapetrous carotid canal, despite the presence of the canal; the presence of stenosis at the site of entry of the artery in the tympanic cavity; the existence of pharyngeal branches arising from the same vessel; the possible occurrence of an occipital artery also arising from the same vessel; the

fact that during surgery this artery (excluding the tympanic part of its course) was mistaken in the cervical region for an authentic ICA since it ran through the same region as the ascending pharyngeal artery.

On the basis of this description, a similar analysis can be used to demonstrate that the anomalies of cerebral vascularization related to an uncommon course of the ICA [2, 5, 6, 7, 9, 12] correspond in fact to segmental agenesis of the ICA. Data from fundamental embryology [4, 13] allow to propose the following scheme of development of the ICA.

At an initial stage of development (Fig. 6A) the ventral aorta (VA) and dorsal aorta (DA) are anastomosed by a certain number of arterial structures, the aortic arches. The arches are numbered from 1 to 4 in the craniocaudal direction. Other embryonic structures, also present at this stage are the primitive maxillary artery (MI), dorsal ophthalmic artery (OD), ventral ophthalmic artery (OV), anterior cerebral artery (CBA) and longitudinal neural system (LNS).

In the interval between this and the subsequent stages certain regions undergo modification via arterial regression (circled arrows), i.e. in the regions of the ventral ophthalmic artery, dorsal aorta and ventral portion of the first two aortic arches.

At a later stage of development (Fig. 6B) the carotid system takes on a different aspect. The dorsal aorta gives rise to the following structures: the third aortic arch which joins anteriorly to the remnant of the ventral aorta, the latter being reduced to the ventral pharyngeal artery (*ph. vent.*) (future external carotid artery); the hyoid artery (Hy), of which the main collateral, the stapedia artery (*st*), gives rise to the internal maxillary artery when it is annexed by the embryonic external carotid system; the mandibular artery (*m*), a dorsal remnant of the first aortic arch; the primitive maxillary artery which during the earlier stage of development has become the posteroinferior hypophyseal artery (*Hp*). The ventral ophthalmic artery, arising directly from the dorsal aorta, develops into the primitive ophthalmic artery (*OI*).

The regression of two important structures now occurs (circled arrows), i.e. the proximal part of the stapedia artery and the portion of the dorsal aorta lying above the third aortic arch.

At an even later stage of development (Fig. 6C) the carotid system shows a practically classical arrangement. The ventral cephalic aorta has become the common carotid artery at this stage and is divided into two branches, the internal carotid (*c. int.*) and external carotid (*c. ext.*) arteries. The ven-

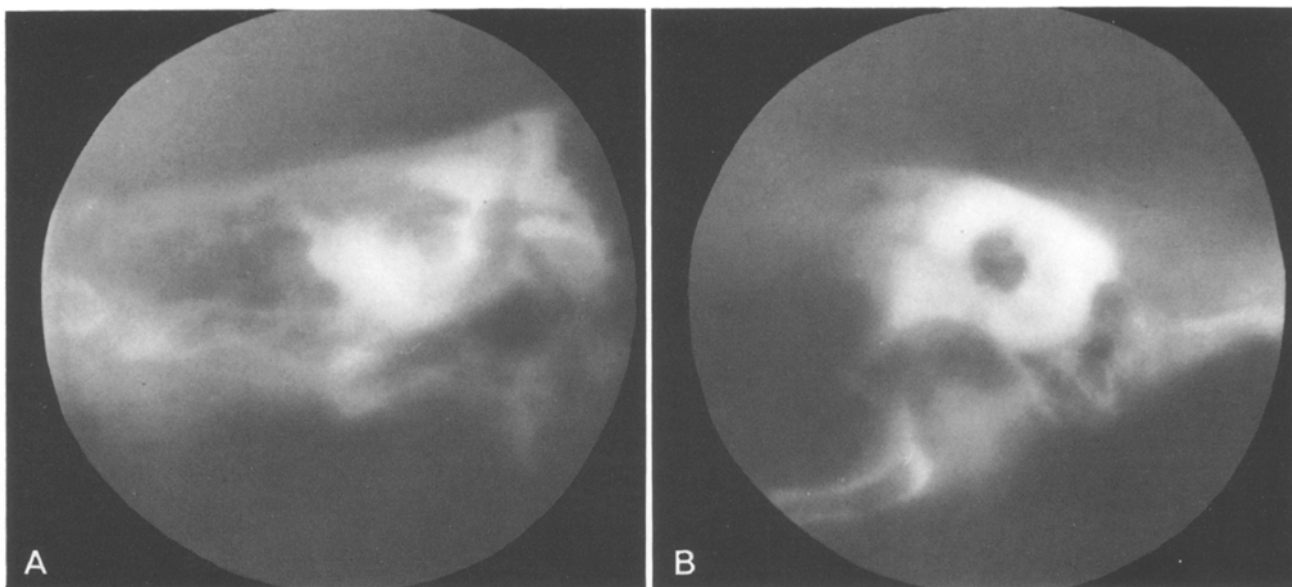


Fig. 5A, B

Tomograms (**A** Stenver's incidence, **B** lateral view) demonstrating the congenital origin of the agenesis since no carotid canal can be seen in the petrous bone

Les tomographies en incidence de Stenvers (**A**) et de profil (**B**) confirment la nature congénitale de l'agénésie en ne mettant pas en évidence de canal carotidien dans la pyramide pétreuse

tral pharyngeal system has by now developed into the faciolingual system (FL), whereas the stapedia system has given rise to the internal maxillary (m. int.) and middle meningeal (mm) arteries. The proximal part of the hyostapedial trunk gives rise to the caroticotympanic artery (CT). The mandibular and posterior hypophyseal arteries have been described above. The inferolateral trunk (Til) (or intracavernous collateral of the internal carotid artery) is a remnant of the dorsal ophthalmic artery, while at this stage the primitive ophthalmic artery has become the definitive ophthalmic artery (OPH) through complex developmental stages whose description is beyond the scope of this paper.

The different segments of the fully developed internal carotid system (Fig. 6D) correspond to the following embryonic structures: the third aortic arch (segment 1) and dorsal aorta between the second and third aortic arches (segment 2); the dorsal aorta between the first and second aortic arches (segment 3); the dorsal aorta between the first aortic arch and primitive maxillary artery (segment 4); the dorsal aorta between the primitive maxillary artery and inferolateral trunk (segment 5); the dorsal aorta between the inferolateral trunk and its terminal branches (segment 6).

The seventh, terminal segment of the internal carotid artery, lying between the primitive (and finally definitive) ophthalmic artery and anterior ce-

rebral artery, is not discussed herein since it is part of another system of arterial compensation (including notably the circle of Willis).

The different embryonic parts of the internal carotid artery can be described with respect to traditional bony anatomical landmarks. These different segments (Fig. 6E), from the origin to the termination of the ICA, are the following: 1) the cervical segment; 2) the initial ascending intrapetrous segment; 3) the distal horizontal intrapetrous segment; 4) the segment ascending in the sphenoid fissure (SF) and through the cavernous sinus; 5) the horizontal segment of the carotid flexure, and finally; 6) the clinoid segment.

These segments appear to be autonomous from the embryological standpoint since each one of them may show segmental agenesis. Furthermore, each segment lies between the origins of two consecutive embryonic vessels. The position of the latter is known in adults since these embryonic vessels may persist in rare, nonpathological cases (Fig. 6F). These embryological vessels include: the hyoid artery and its stapedia branch between segments 2 and 3 of the ICA; the otic artery between segments 3 and 4; the trigeminal and primitive maxillary arteries, between segments 4 and 5; the dorsal ophthalmic artery, between segments 5 and 6; the ventral ophthalmic artery distal to segment 6.

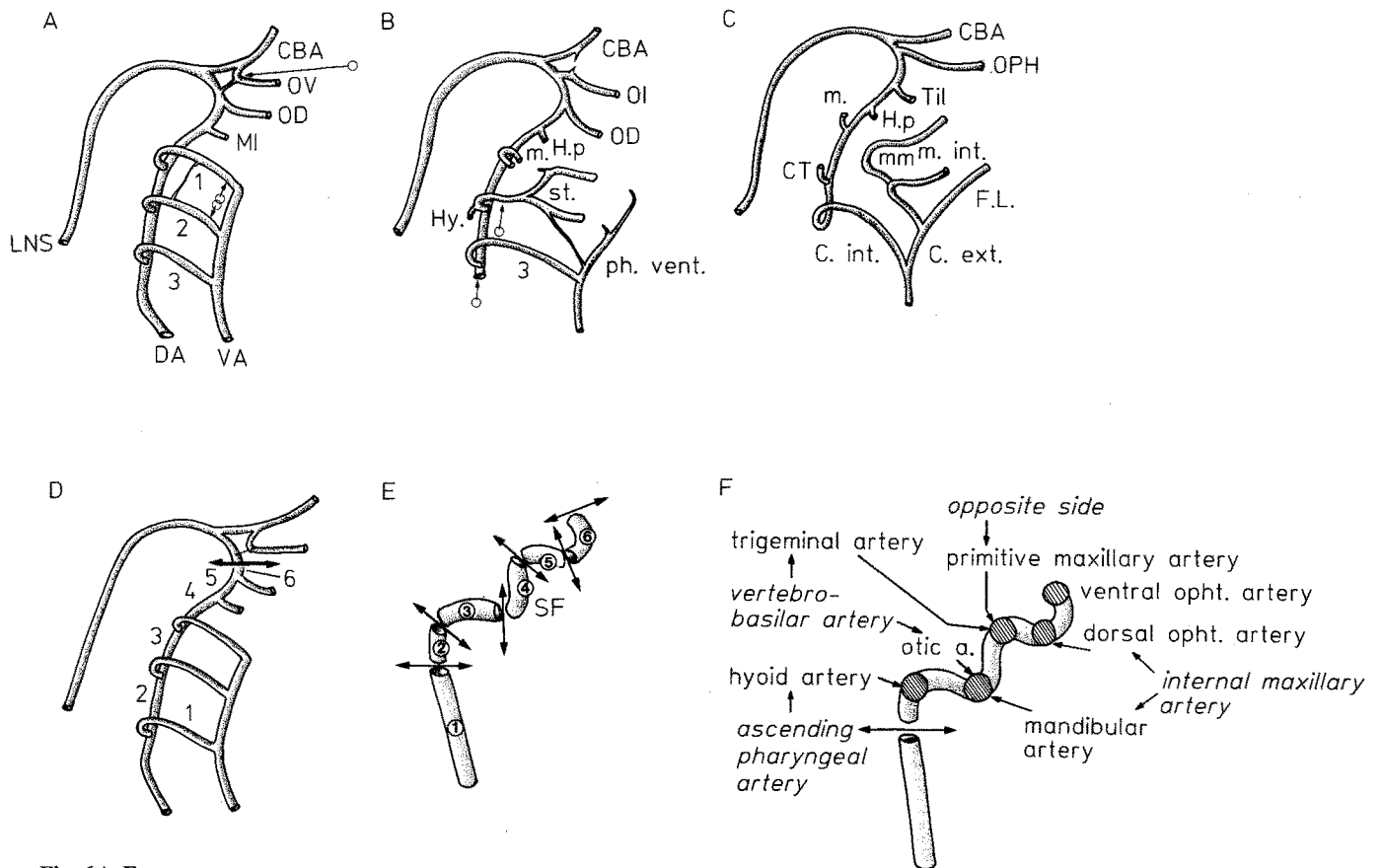


Fig. 6A-F

Illustration of the embryonic development of the aortic arches in the cephalic region. Successive stages of development are shown in **A**, **B** and **C**. **D**, **E** and **F** highlight the specific development of the internal carotid artery, its different segments (**E**) and the embryonic vessels and their origin (**F**)

A 1, 2, 3 Aortic arches numbered in the craniocaudal direction. *CBA* anterior cerebral artery, *OV* ventral ophthalmic artery, *OD* dorsal ophthalmic artery, *MI* primitive maxillary artery, *LNS* longitudinal neural system, *DA* dorsal aorta, *VA* ventral aorta. **B** *OI* primitive ophthalmic artery, *st* stapedial artery, *Hy* hyoid artery, *ph. vent.* ventral pharyngeal artery. **C** *OPH* Definitive ophthalmic artery, *Til* intracavernous collateral of the internal carotid artery, *mm* middle meningeal arteries, *CT* caroticotympanic artery, *m.int.* internal maxillary artery, *FL* faciolingual system, *C.int.* internal carotid, *C.ext.* external carotid artery. **D** 1 Third aortic arch, 2 dorsal aorta between the second and third aortic arches, 3 dorsal aorta between the first and second aortic arches, 4 dorsal aorta between the first aortic arch and the primitive maxillary artery, 5 dorsal aorta between the primitive maxillary artery and the inferolateral trunk, 6 dorsal aorta between the inferolateral trunk and its terminal branches. **E** 1 Cervical segment, 2 initial ascending intrapetrous segment, 3 distal horizontal intrapetrous segment, 4 segment ascending in the sphenoid fissure (*SF*) and through the cavernous sinus, 5 horizontal segment of the carotid flexure, 6 clinoid segment.

Représentation schématique du développement embryonnaire des arcs aortiques dans la région céphalique. Successivement **A**, **B** et **C**. **D**, **E** et **F** soulignent le développement spécifique de la carotide interne, ses différentes portions (**E**) et les différents vaisseaux embryonnaires et leur tronc artériel d'origine (**F**) (abréviations: voir légende anglaise)

In the adult, each of these embryonic vessels may be the point of entry of vascular rerouting in cases of segmental agenesis of part of the ICA proximal to the embryonic vessel (Table 1). The following types of vascular rerouting can be identified: cervical agenesis – rerouting via the hyoid artery through the ascending pharyngeal artery; cervical and petrous agenesis – compensation by the vertebro-basilar system via the otic or trigeminal artery as in our patient; cases of more advanced agenesis – via the primitive maxillary

artery originating from the carotid flexure on the contralateral side [5, 8, 9]; combined cervical, petrous and cavernous agenesis – rerouting through a complex cavernous network leading to the internal maxillary artery.

In cases of segmental agenesis it seems that the embryonic vessel immediately distal to the missing segment is incapable of constituting a sufficient entry point for vascular rerouting. Accordingly, the next (more distal) embryonic vessel becomes the source of compensatory arterial flow ensuring

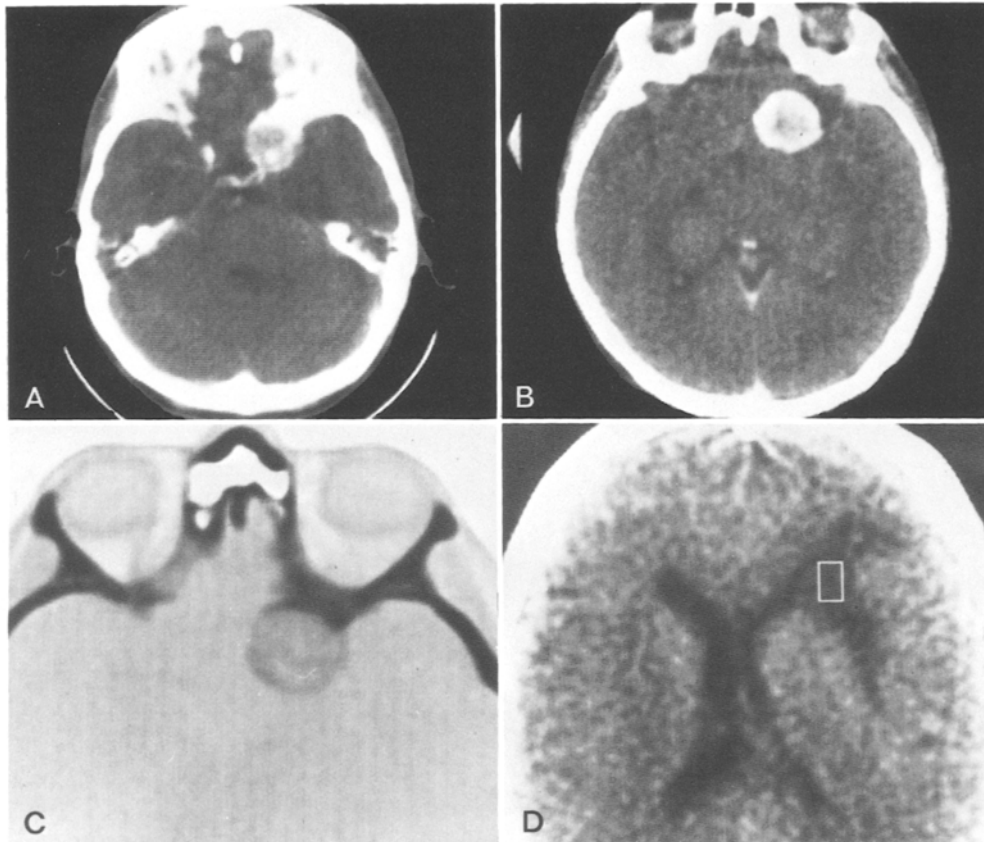


Fig. 7A-D

CT scans of the patient are described in the paper. Although the lesion appears to be inserted on the roof of the orbit and anterior clinoid process (A, C), the upward extension (B) and hemispheric hypodensity (D) suggested a preliminary diagnosis of metastatic tumor rather than meningioma

Explorations scanographiques. Bien que la lésion semble insérée sur le toit de l'orbite et la clinioïde antérieure (A et C), l'extension vers le haut (B) et l'hypodensité hémisphérique (D) ne font pas envisagées en premier le diagnostic de méningiome mais plutôt celui de métastase

vascularization of the deficient internal carotid territory. For example, in cases of agenesis of the petrous segment of the ICA the vertebral anastomosis via the otic artery does not persist and thus the trigeminal artery becomes the source of vascular rerouting. Likewise, in cases of agenesis of the vertical part of the carotid flexure, rerouting via the contralateral carotid flexure and primitive maxillary artery (or a remnant of the latter or of the trigeminal artery) is not possible. In such cases, a cavernous compensatory network supplied by the internal maxillary artery (corresponding to a remnant of the dorsal ophthalmic artery) ensures internal carotid function. Finally, data from the literature and the observations in our patient seem to indicate that agenesis of one segment of the ICA leads to the regression of all segments proximal to the anomaly. In our patient agenesis of the horizontal intrapetrous segment led to retro-

grade agenesis of the ascending petrous and cervical segments.

It is conceivable that such proximal regression occurs via a mechanism similar to that which produces small carotid vessels proximal to the site of acquired stenosis of the intracavernous flexure of the ICA.

In sum, it can be seen that the ICA is constituted by the persistence of embryologically different arterial segments, each of which may be the site of segmental agenesis. Knowledge of these embryological findings allows to identify the type of agenesis and the mode of compensatory vascular rerouting to the remaining segments of the ICA. However, although many varieties of segmental agenesis are described in this paper there have not been to our knowledge any reports in the neuro-radiological or anatomical literature describing revascularization via the otic, mandibular or trigem-

Table 1. Congenital anomalies of the trunk of the internal carotid artery (see also Fig. 6E)

Abnormal embryonic segment	Type of carotid anomaly	Vascular rerouting
3rd aortic arch (segment 1)	Fenestration of cervical segment Agenesis of cervical segment	Ascending pharyngeal artery (intratympanic part of ICA) Point of entry: junction of segments 2-3
Dorsal aorta between 2nd and 3rd aortic arches (segment 2)	Agenesis of cervical segment and ascending intrapetrous segment	Otic artery Point of entry: junction of segments 3-4
Dorsal aorta between 1st and 2nd aortic arches (segment 3)	Agenesis of cervical and petrous segments	Trigeminal artery and contralateral carotid flexure Point of entry: junction of segments 4-5
Dorsal aorta between 1st aortic arch and dorsal ophthalmic artery (segment 4)	Agenesis of cervical, petrous and cavernous segments	Cavernous arterial network Point of entry: junction of segments 5-6

inal artery. The patient presented in this paper would thus be the first reported case of such vascular rerouting.

Conclusion

It is clearly established that the hemodynamic response which occurs to compensate an agenetic arterial segment corresponds to a stable and classical anatomical arrangement in a given species [3]. However, the following remark should be made, as pointed out by Vera [16] who quoted Lazorthes and Gouazé [11] in his doctoral thesis: "The cavernous arterial network in man in no way corresponds to the persistence of an embryological pattern, but rather represent a developmental shift towards a pattern found in certain mammalian species inferior to the Primates". In our opinion, this remark on the cavernous circulation is applicable to all arterial patterns and in particular, agenesis of the internal carotid artery. Finally, although the scheme presented in this paper is explicative and apparently intellectually satisfactory, it lacks many details and may contain certain errors, even though all previously described anatomical variations of the internal carotid artery have to our knowledge been incorporated in it. Indeed, although it may be practical to consider the embryonic vessels as tubal structures, it is well known that the arterial

system is initially a reticular structure prior to the development of specialized vascular channels, the arteries.

A similar analysis can be used to describe the heterogeneous embryological origin of the vertebral artery [9] and to allow understanding of the "normal" anatomical variations of this vessel in the upper cervical region.

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