Stroke Associated with Elongation and Kinking of the Internal Carotid Artery:

Report of Three Cases Treated by Segmental Resection of the Carotid Artery *

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ALTHOUCH the carotid syndrome is reported to have been described by Chiari over 50 years ago and reaffirmed in this country by Hunt in 1914,1 general interest has been stimulated only in the recent few vears by the application of surgical technics to the correction of the cervical carotid stenosis responsible for the symptoms of hemiparesis. Eastcott, Pickering and Rob⁴ reported successful excision of the narrowed carotid bifurcation with end-to-end anastomosis of the common to the internal carotid artery in 1954, Cooley, Al-Naaman and Carton¹ in 1956 first reported successful thromboendarterectomy of the region of the carotid bifurcation, making use of a temporary shunt to maintain flow during the period of occlusion. Strulley, Hurwitt and Blankenberg⁷ had previously attempted endarterectomy, but had met with failure because of extension of organized thrombus up into the skull. Lin, Javid and Doyle⁵ have reported resection of a partially occluded internal carotid artery and replacement with a vein graft. Finally Denman, Ehni and Duty ³ reported the use of arterial homograft to bridge the defect between the common and internal carotid created by resection of an atheromatous stenosis. Now an ever increasing number of case reports and small series is appearing in the literature in testimony of the generalized interest in this syndrome and its possible surgical correction. To the best of our knowledge, in all reported instances the obstruction to the carotid circulation has been on the basis of intimal thickening due to atherosclerotic deposits at or near the carotid bifurcation with or without secondary thrombosis. The following cases, however, demonstrate an unusual form of internal carotid occlusion not secondary to intrinsic narrowing of the lumen, but simply due to a remarkable elongation of the vessel of sufficient degree to produce narrowing by way of kinking with associated transient hemiparesis. Having been previously unaware of such a condition and being unable to find a report of such in the literature, we report the following cases.

Case Reports

Case 1. A 69-nine-year-old white female with arterial hypertension in the general range of 230 over 110 mm. Hg had been followed for several years. A number of antihypertensive drugs had been administered without good results, usually because of subjective side effects to their administration. In association with her hypertension, she had for a like number of years complained of pain in the left occipitoparietal region of the scalp. On April 29, 1958, she noted numbress of the whole right side of the body with concomitant weakness of such degree that she was unable to walk without assistance, the right leg giving way under her. When seen the following day her blood pressure was 235 over 115, and there was gross muscular weakness of the extremities of the right side of the body without reflex changes. The temporal and carotid pulses were palpable bilaterally. Because of persistence of the symptoms of hemiparesis, she was admitted to the Telfair Hospital five days later

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and a percutaneous carotid arteriogram was made (Fig. 1). The initial injection of 10 cc. of 50 per cent Hypaque extravasated into the soft tissues surrounding the carotid artery, somewhat obscuring the region of the carotid bifurcation on the second successful injection, which is shown. The internal carotid artery was seen to pass posteriorly and upward and then angulate sharply forward and downward before ascending into the carotid canal of the temporal bone, with apparent rather marked constriction of the lumen at the point of angulation. During the following week the patient's numbness and motor weakness of the right side subsided. She was transferred to the Oglethorpe Sanatorium where, on May 12, 1958, under general anesthesia she was submitted to exploration of the left carotid artery. As the vessel was dissected out it was seen that there did exist a kinking of the vessel upon itself of such a degree that pulsations in the vessel distal to the kink were markedly diminished (Fig. 2). The level of the carotid bifurcation was considerably higher than usual indicating that the process was involving not merely the internal carotid, but certainly the common carotid artery and likely the arch of the aorta as well. The elongation was of such a degree that



FIG 1. Percutaneous left carotid arteriogram showing elongation and kinking of the internal carotid artery. A previous injection had resulted in extravasation into the soft tissues around the artery. (Case 1.)



FIG. 2. The kink in the internal carotid artery is shown (arrow). The level of the kink in the artery could be varied by manipulation, but the kink remained severe in all positions. (Case 1.)



FIG. 3. After resection of a segment of the common carotid artery, which is lying alongside the point of anastomosis, the internal carotid now arches gently toward the base of the skull. (Case 1.)

it was not considered feasible to pull the bifurcation down, although some thought was given to the use of fascia slings or tacking sutures to accomplish this. When clamps were applied to the common carotid artery, a faint pulse could be felt in the external and internal carotid vessels distally. Apparently collateral circulation by way of the external carotid was sufficient to provide reasonable flow. It was considered that actual shortening of the vessel by resecting a portion of the common carotid artery offered a relatively safe and, at the same time, sure way to correct the abnormality. The common carotid artery was cross-clamped low in the neck and again just below the bifurcation, and a segment measuring two centimeters in length after removal was resected. End-to-end anastomosis was carried out with continuous #00000 arterial silk. The total time of occlusion was thirteen and one-half minutes. This served to relieve the kinking of the internal carotid artery completely, allowing it to arch gently upward toward the base of the skull (Fig. 3). A strong pulse was now palpable in the vessels distal to the anastomosis. Recovery from anesthesia was prompt, and the postoperative course was uneventful. The only

demonstrable neurologic deficit was the subjective complaint of numbness of the fingers of the right hand. Subsequent follow up has shown no further episodes similar to that of her chief complaint, and there has been gradual improvement of the numbness in the fingers of the right hand. Her hypertension has remained essentially as before. Postoperative arteriogram (Fig. 4) has shown complete relief of the kinking of the carotid artery.

Case 2. A 75-year-old white man was admitted to the Memorial Hospital of Chatham County on September 4, 1958, with a history of several years of intermittent dizzy spells, and a very transient episode of slight weakness of the right side two months prior to his admission. On the morning of admission as he leaned over to tie his shoes he found that the right side was numb and weak. Within a matter of 30 minutes he developed complete hemiparesis on the right side and complete aphasia, which were confirmed by examination by one of us. By the time of admission to the hospital some four hours after the onset, the hemiparesis had partially cleared, leaving him with a slightly weak, ataxic right side. He was now able

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to talk with some difficulty. His past history was positive for the gradual development of a central blindness due to optic atrophy over a period of several years. He had a chronic cough for many years. On physical examination, the blood pressure was 130/70. Temperature, pulse, and respiration were normal. There was motor-weakness of the right upper and lower extremities, worse in the upper extremity, and with gross ataxia on voluntary motion. The remainder of the physical examination was not remarkable. There were no overt reflex changes. The spinal tap shortly after admission revealed crystal clear spinal fluid under normal pressure. Bilateral carotid arteriogram was carried out within the next few hours (Fig. 5). Film on the right side showed a rather corkscrew type enlongation of the internal carotid just below the skull, while on the left side there was a suggestion of a plaque at the carotid bifuration which was not well seen. In addition, about two centimeters above the bifurcation there was noted a band-like stricture of the internal carotid artery, the exact significance of which was not appreciated. Because of the uncertainty of the vessel changes on the left side, exploration of the left carotid artery under local anesthesia was advised and carried out on September 5, 1958, the day following admission. At exploration, there was found to be a nonobstructing atheromatous plaque in the common carotid artery.



FIG. 4. Postoperative arteriogram of Case 1. The kink has effectively been straightened and the common carotid anastomosis is widely patent.



FIG. 5. Percutaneous arteriogram of the right carotid artery, A (left), shows a corkscrew deformity just below entry into the skull. Injection of the left side, B (right), demonstrates elongation and a suggestive transverse radiolucent band in the internal carotid artery (arrow). (Case 2.)



Case 3. A 68-year-old white male was admitted to the Oglethorpe Sanatorium October 11, 1958, with a history of having developed profound weakness of the right upper extremity during the night four days prior to admission. He had awakened during the night to note that his arm was weak. The following morning he found that it was completely paralyzed and that he was unable to speak any word except "no." There was no noticeable involvement of the right leg. He remained about the same for 48 hours following which slow progressive improvement began. At the time of his



FIG. 7. Postoperative left carotid arteriogram (Case 2). The elongation has been reduced and the vessel including the anastomosis is widely patent. The external carotid was ligated at surgery.



FIG. 6. Photograph of the left carotid bifurcation and internal carotid artery at operation (Case 2). Kinking of the internal carotid artery varied in severity depending on manipulation of the vessel.

There was, however, elongation and kinking of the internal carotid, with diminution of the pulse distal to the kink (Fig. 6). Because the plaque at the carotid bifurcation would not have had to enlarge greatly to produce significant internal carotid obstruction, it was felt wisest to try to treat the plaque and the kink at the same time if possible. Being reminded of the method used by Eastcott et al.² in treating their first case of the carotid syndrome, it was elected to resect the carotid bifurcation and carry out anastomosis between the end of the common carotid and the end of the internal carotid. The common and external carotid arteries were occluded with arterial forceps for two and one half minutes as a test, and no subjective or objective neurologic change was evident. The patient was able to converse quite normally. Accordingly the carotid bifurcation was resected, and end-to-end anastomosis was carried out with #00000 arterial silk. The total occlusion time was eight minutes and 30 seconds. During the latter part of the occlusion, the patient became quite groggy and lethargic and was unable to answer questions intelligently. However, when the clamps were removed and a good pulsatile flow reestablished through the carotid artery, he immediately became alert again and was able to converse normally during the closure of the wound. His postoperative course was completely uneventful, and

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FIG. 8. A (left), Percutaneous right carotid arteriogram (Case 3) shows tortuosity of the internal carotid with a suggestion of kinking just above the bifurcation. B (right), Arteriogram of the left carotid shows marked elongation without kinking at the time of injection.

admission to the hospital there was marked weakness of the right upper extremity. There was considerable hesitancy of speech and slurring of words. There were no reflex changes. The family history was positive in that one brother has had three strokes and is still living. Spinal tap was performed shortly after his admission, and produced crystalclear spinal fluid under normal pressure. Bilateral carotid arteriogram was also carried out on the day of admission after the spinal tap. On both sides there was seen to be marked elongation of the carotid arteries (Fig. 8). No actual kink was noted. A Matas test was performed the following day, compressing the left carotid artery for six minutes, during which time there were no subjective or objective changes. In view of our previous experience and because of the completely normal appearance of the intracerebral vessels exploration of the left carotid was advised. On October 14, 1958, under local anesthesia, this exploration was carried out. The remarkable elongation of the carotid artery was confirmed. It was interesting that one could manipulate the carotid into various forms of bending and twisting, so that at times the flow might completely kink off, while in other positions simply a remarkable tortuosity would result. Figure 9 serves to illustrate these points. An atheromatous plaque could be palpated in the com-

mon carotid artery just proximal to the bifurcation. This was small and not occluding either the internal carotid or the external carotid branch. Using the same basic reasoning as in case two, it was elected to attempt to resect the carotid bifurcation. A four-minute test occlusion of the common carotid and external carotid arteries brought no objective or subjective change on the part of the patient. After a brief period of rest the vessels were reoccluded, and the carotid bifurcation was resected. removing the total length of 2.8 cm. of common and internal carotid artery. End-to-end anastomosis was carried out between the common carotid artery and the relatively small internal carotid. Due to the disproportionate size, the anastomosis was rather difficult, and the total occlusion time was 17 minutes. During the occlusion the patient did develop numbness of the right upper extremity. There was no change in his state of consciousness, however. The external carotid artery was simply ligated. After removal of the occluding clamps a strong pulse was palpable in the distal internal carotid artery. There was no longer any tortuosity. This was confirmed by repeat arteriogram made one week after operation. The postoperative course was marked by significant recurrence of weakness in the right upper extremity and increased slurring of the speech on the night of operation. The fol-



Fig. 9. Photographs of the left internal carotid taken at operation demonstrates the variability of position and consequently the severity of kinking which can be produced. On the left (A) the vessel doubles back on itself and then ascends out of sight. This is how it was found at surgery. On the right (B) after opening the carotid sheath, a severe kink is able to be produced.

lowing day, however, the patient had regained his preoperative status and has subsequently shown progressive improvement so that two weeks postoperatively only the very slightest hesitation in speaking some of his words, and perhaps slight ataxia in the right upper extremity are present.

Discussion

Although internal carotid occlusion secondary to kinking from elongation of the type reported above has apparently been previously unreported, it would seem that with the increasing investigation of the carotid arterial tree, more of these will be encountered. Similar elongation of the carotid artery presenting as an aneurysmoid pulsation in the neck, but without kinking was seen several months prior to the above cases and evaluated by arteriograms (Fig. 10). In the absence of symptoms no surgical attack was performed on this case and the patient is being followed with the anticipation that he might develop a similar condition in the future.

Unfortunately, a right carotid arteriogram was not made in the first patient. In the subsequent two patients there was evidence by arteriogram of elongation and tortuosity of both carotid arteries. In view of the fact that significant kinking has either existed or been relatively easy to produce in each of these patients at the time of operation, the possibility arises that it may require simultaneous kinking of both sides to produce the stroke in the absence of significant intracranial arteriosclerosis. This is likely in view of the evidence of significant collateral circulation at the time of operation in all of these patients.

The principal decision which had to be made in our first case was whether to proceed with carotid occlusion without a shunt in the absence of hypothermia. The decision for doing so in this case was predicated on the evidence of good collateral flow in the carotid distally. We were aware that Bahnson had occluded first the innominate



FIG. 10. Asymptomatic case of severe internal carotid tortuosity. No surgery has been performed in view of absence of symptoms.

and subsequently the left common carotid artery for periods approaching 20 minutes each in replacing an aortic arch aneurysm with homograft. This patient had recovered promptly without evidence of neurologic damage. On the other hand, in carrying out a similar procedure Cooley ² reported cerebral damage from ischemia resulting from thrombosis of a carotid shunt even in the presence of hypothermia. Rob and Wheeler ⁶ have reported transient hemiplegia following seventeen minutes of occlusion of the carotid artery at normal temperatures. The variability of collateral circulation, and its importance in the prevention of permanent neurologic sequels, can be found in the patients with atheromatous narrowing of the internal carotid at its origin. We have seen patients with total occlusion of the vessel with no symptoms on the one hand, and we have seen fatal stroke due to partial occlusion on the other hand. It would seem, on the basis of our experience here, that the presence of a pulse distally after occlusion

of the common carotid is a reliable indicator of good collateral circulation. We feel, however, that hypothermia should be available and should be applied if exploration indicates an inadequate collateral flow.

In cases two and three we were able to carry out a test period of occlusion and establish the safety of occlusion in advance because the patients were under local anesthesia. It is of interest that in both these it was necessary to occlude completely the collateral flow through the external carotid artery. It is also of interest that the pulse in the internal carotid above the point of occlusion in both cases was completely obliterated by the occlusion. This would, we believe, tend to strengthen the earlier contention that the presence of a pulse in the vessel distally after occlusion would be a reliable indicator of adequate collateral flow.

At the operating table in each of these patients, we were impressed with the flexibility of the arteries. Aside from the localized plaque in the carotid bifurcation in two cases there was no significant sclerosis of the vessels. Ouite obviously the process is one of elongation due to loss of elasticity. As the vessel elongates it bows on the fixed points at the base of the skull and thoracic inlet. The bowing is limited by the resistance of the fascia of the carotid sheath. Then when the position of the head is right the vessel is thrown into a kink. As was demonstrated at the operating table in these patients there is a much greater tendency to kinking as the head is turned toward the side of the vessel in question. This has been confirmed by arteriograms in case three which are not shown.

Not generally appreciated, we believe, is the facility with which operation of this type can be done under local anesthesia. Dissection and clamping of the vessels is well tolerated. The patients do complain quite bitterly if penicillin solution is instilled into the wound. We believe that local anesthesia will gain wider acceptance for initial exploration and allow test periods of occlusion before proceeding to general anesthesia with hypothermia when necessary.

Summary

Three cases of partial internal carotid artery occlusion on the basis of degenerative elongation of the carotid system with kinking of the internal carotid are presented. One patient was treated by segmental resection of the common carotid artery with endto-end anastomosis with a good result. The other two patients, because of the presence of a partially stenosing atheromatous plaque in the carotid bifurcation, were treated by resection of the carotid bifurcation with end-to-end anastomosis of the common ca-

rotid to the internal carotid artery. A fourth case of similar elongation of the carotid artery without symptoms from kinking is reported. It is felt that more cases will be forthcoming as the result of increasing angiographic investigation of the carotid artery currently in vogue. Occlusion times of thirteen and one-half minutes of the common carotid artery, and of eight and one-half minutes and 17 minutes of the internal carotid artery were tolerated without serious ill effects. Some of the factors relating thereto are discussed. Local anesthesia is recommended for preliminary exploration before proceeding to general anesthesia with hypothermia if necessary.

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