Seven Cerebral Aneurysms: A Challenging Case from the Andean Slopes Managed with 1-Stage Surgery

Joham Choque-Velasquez[1,2], Roberto Colasanti[1-3], George Fotakopoulos[2], Humberto Elera-Florez[2], Juha Hernesniemi[1,2]

■ BACKGROUND: **Treatment of multiple intracranial aneurysms is particularly demanding and even more so in a developing country where access to specialized centers may be prevented by different factors.**

■ METHODS: **Single-stage surgical treatment of 7 cerebral aneurysms was performed in a 58-year-old woman from the northern Peruvian Andes.**

■ RESULTS: **All 7 aneurysms were successfully and safely clipped through 2 lateral supraorbital craniotomies. The double clip technique was used in 3 aneurysms to prevent any residual aneurysmal neck.**

■ CONCLUSIONS: **Good teamwork and correct application of microsurgical principles may allow effective treatment in complex neurosurgical cases even in resource-challenged environments.**

INTRODUCTION

Management of multiple intracranial aneurysms is particularly demanding and even more so in a developing country where access to specialized centers may be impeded by different factors. Economic issues and an inequitable geographic distribution of major specialized services, together with the presence of ancestral indigenous health beliefs, contribute to exacerbate the disparities in health care quality between rural and urban populations in Perù.[1-3] The transfer system of a neurosurgical patient from a remote region to one of the few highly specialized centers in the coastal areas may be dangerous as well as an economic burden.

In 2013, EsSalud (Social Security Health Care System of Peru) initiated a plan called "Plan confianza," with the aim to improve the aforementioned problems. This project promotes the development of specialized neurosurgical centers in major strategic places all around the country to make optimal treatment of neurosurgical patients feasible. The creation of a Neurosurgical Center of Excellence in Trujillo, led by Professor Juha Hernesniemi and his team, falls within this plan. We present a challenging case that was managed in the new Neurosurgical Center of Trujillo involving the single-stage surgical treatment of 7 cerebral aneurysms in a 58-year-old woman from the northern Peruvian Andes.

CASE REPORT

A 58-year-old Andean woman living in Perù at ≥3000 m above sea level was admitted to her local hospital in Cajamarca with symptoms of severe headaches, dizziness, neck pain, and photophobia, all progressive over the past several weeks. Computed tomography (CT) scan showed an acute subarachnoid hemorrhage (Fisher grade 3). Because of the difficulties in being transported to a hospital with a neurosurgical unit, she was initially managed with conservative treatment.

After 3 weeks of conservative treatment and following travel of >5 hours, she was transferred to our recently opened neurosurgical center in Trujillo. On clinical examination, she was neurologically intact (World Federation of Neurosurgical Societies [WFNS] grade I). Her pathologic anamnesis was unremarkable, and she had no family history of cerebral aneurysms. CT angiography with three-dimensional reconstructions demonstrated the presence of 7 aneurysms arising from the 1) right anterior choroidal artery, 2) right posterior communicating artery, 3) A1

Key words
- Developing countries
- Lateral supraorbital approach
- Microneurosurgery
- Mirror aneurysms
- Multiple intracranial aneurysms
- Single-stage management
- Subarachnoid hemorrhage

Abbreviations and Acronyms
CT: Computed tomography
WFNS: World Federation of Neurosurgical Societies

From the [1]Department of Neurosurgery, Helsinki University Central Hospital, Helsinki, Finland; [2]Neurosurgical Unit, EsSalud La Libertad, Trujillo, Peru; and [3]Department of Neurosurgery, Umberto I General Hospital, Università Politecnica delle Marche, Ancona, Italy

To whom correspondence should be addressed: Joham Choque-Velasquez, M.D. [E-mail: johchove@hotmail.com]

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segment of the right anterior cerebral artery, 4) bifurcation of the right middle cerebral artery, 5) left posterior communicating artery, 6) left internal carotid artery, and 7) M1 segment of the left middle cerebral artery (Figure 1). The left posterior communicating aneurysm was thought to have ruptured on the basis of the referred location of the subarachnoid hemorrhage.

After careful evaluation of the radiologic images and taking into consideration that there is no endovascular unit in our center, we decided to attempt a single-stage surgical procedure to treat all 7 aneurysms. The patient was placed supine on the operating table, and her head was secured in a Sugita fixation device. The head was positioned in neutral position and was elevated approximately 20 cm above the level of the heart to facilitate venous drainage. Then 2 lateral supraorbital craniotomies were performed on both sides (Figure 2).

Following both craniotomies, the multiple aneurysms were treated sequentially using standard microneurosurgical technique, starting from the left side. During these stages, the head was rotated 15°–30° toward the opposite side according to the location of the various aneurysms (Figure 3 and Video 1). After opening of the left sylvian fissure and the release of cerebrospinal fluid from the carotid, optic, and chiasmatic cisterns, the 3 aneurysms arising from the left part of the circle of Willis were clipped, beginning with the left posterior communicating artery aneurysm (the one that had bled). Then the head was turned to the left, and the right middle cerebral artery bifurcation aneurysm was clipped after a focused opening of the right sylvian fissure. Finally, the right internal carotid artery was approached, and the 3 remaining aneurysms were subsequently clipped.

Video

Video available at WORLDNEUROSURGERY.org

After brain relaxation was achieved, the general strategy for clipping of aneurysms consisted of the following steps: 1) establishing proximal and distal control of the parent arteries, 2) aneurysm neck dissection under temporary clipping of the arteries, 3) pilot clipping and aneurysm dome remodeling, and 4) definitive clipping and evaluation of the patency of the surrounding arteries. Three aneurysms—the ones originating from the left posterior communicating artery, left internal carotid artery, and M1 segment of the left middle cerebral artery—were clipped by the double clip technique to prevent any residual aneurysmal neck.[4] The wound, including the skin, was closed under the operating microscope. The surgical time skin to skin was 2 hours and 40 minutes, and the microsurgical stage until the clipping of the last aneurysm was 1 hour and 30 minutes.

The patient had an uneventful postoperative course with an intensive care unit stay of only 1 day. Postoperative CT angiography revealed total occlusion of all aneurysms without any complication (Figure 4). The patient continues to do well >6 months after her surgery without any medication. Although she had no family history of cerebral aneurysms, we advised her to undergo genetic analysis and her relatives to undergo screening.

DISCUSSION

Access to specialized health centers in developing countries, especially in rural areas, is usually limited because of economic, cultural, and geographic reasons.[1-3] In Perú, the most developed neurosurgical centers are in the capital city of Lima. The few neuroendovascular units in the country are also concentrated in the capital district. As the patient transfer system does not work effectively, the transport of an inpatient from 1500 km inside the country to Lima is very expensive and may represent a high risk for the patient.

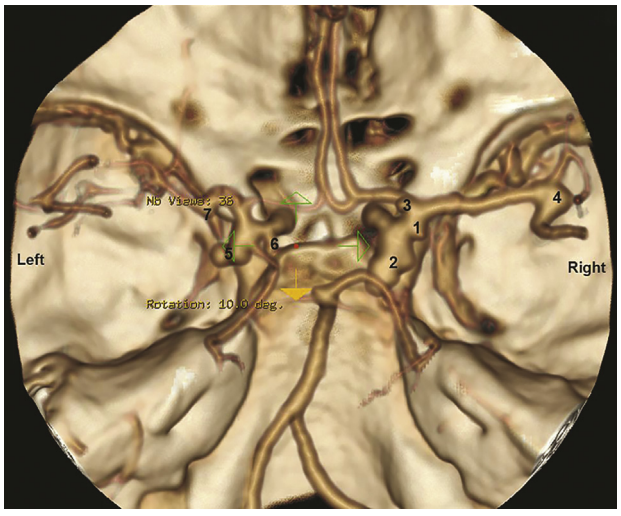

Figure 1. Computed tomography angiography with three-dimensional reconstructions illustrating the presence of 7 aneurysms arising from the right anterior choroidal artery (*1*), right posterior communicating artery (*2*), A1 segment of the right anterior cerebral artery (*3*), bifurcation of the right middle cerebral artery (*4*), left posterior communicating artery (*5*), left internal carotid artery (*6*), and M1 segment of the left middle cerebral artery (*7*).

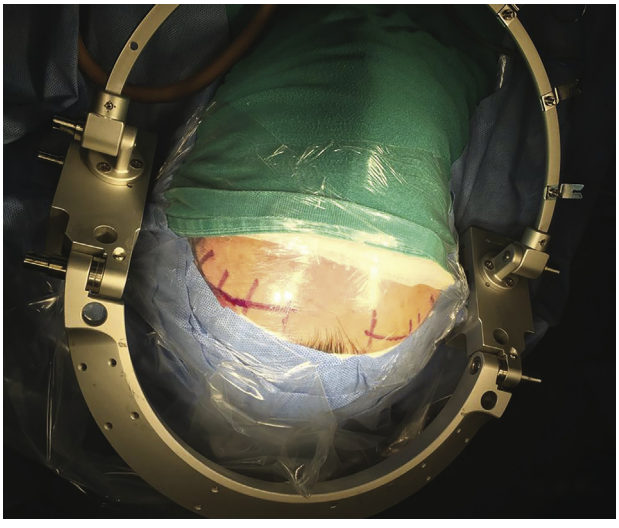

Figure 2. Bilateral lateral supraorbital approach.

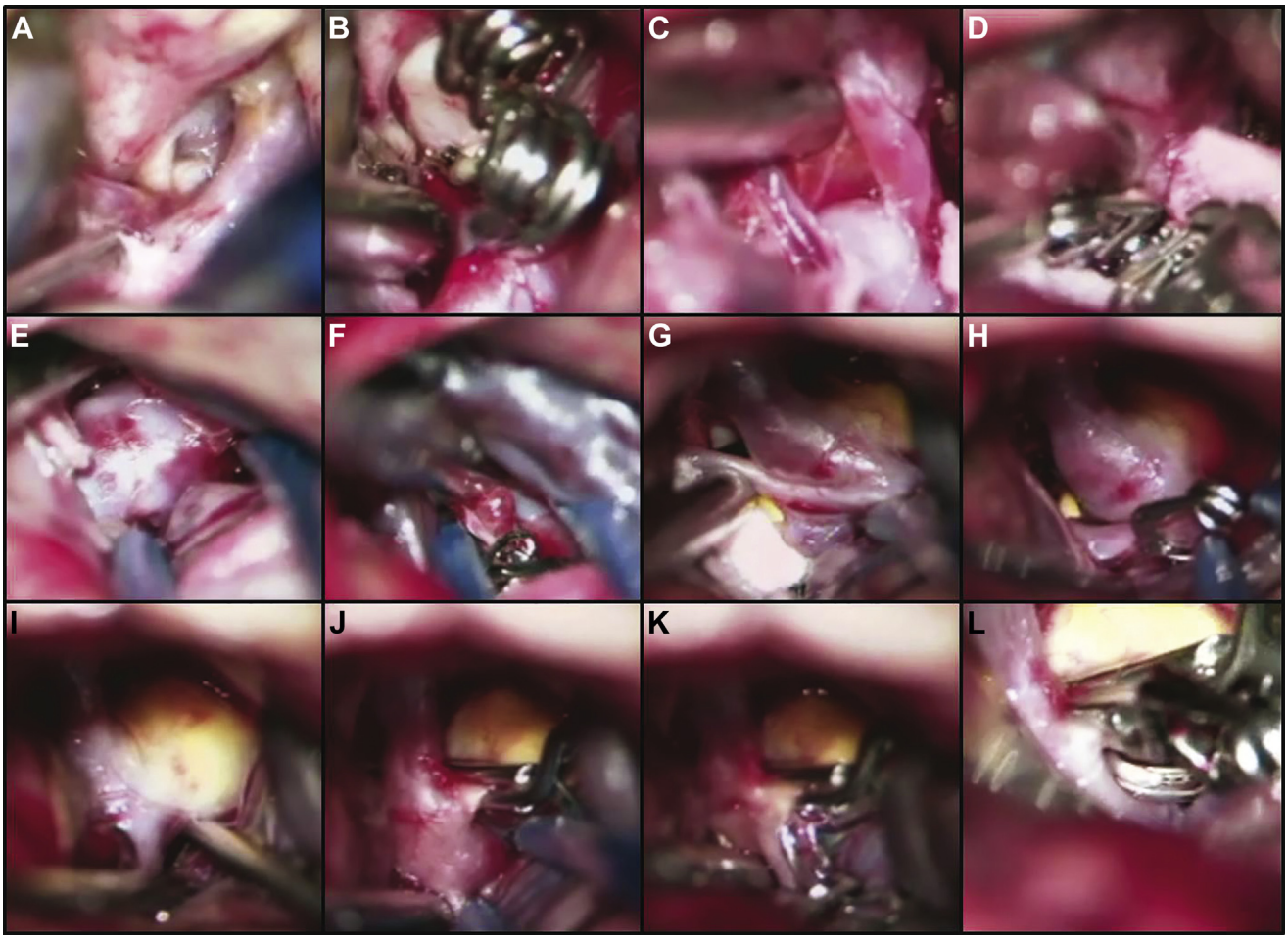

Figure 3. Intraoperative photographs. View of the left posterior communicating artery and internal carotid artery aneurysms (**A**), completely clipped using the double clip technique (**B**). View of the aneurysm of the M1 segment of the left middle cerebral artery (**C**) and of its clipping using the double clip technique (**D**). View of the aneurysm of the right middle cerebral artery bifurcation (**E**) and of its clip occlusion (**F**). View of the aneurysm of the A1 segment of the right anterior cerebral artery (**G**), totally occluded using a straight clip (**H**). The right posterior communicating artery aneurysm (**I**) was occluded with a multiple clipping (**J**–**L**), whereas the right anterior choroidal artery aneurysm (**J**) was occluded using a simple clipping (**K**).

Although there have been several advancements, health system indicators still emphasize considerable unmet clinical need. The shortage of neurosurgeons as well as of neurologists and neuroanesthesiologists is one of the main reasons for this. Moreover, inefficient administration of the resources for neurosurgical care also plays an important role. Many neurosurgical units have inadequate organization. In addition, the limited number of neurosurgical beds (both in neurosurgical wards and in intensive care units) determines an extension of the surgical waitlist.

"Plan confianza" by EsSalud was designed to reduce the inequalities in neurosurgical care between poor and nonpoor patients and between rural and urban populations. We believe that an effective decentralization of major specialized services together with an efficient use of the available resources may help to improve health care systems in developing countries. A judicious decentralization could help to distribute and retain highly specialized health care workers in remote areas, ameliorating treatment outcomes of rural populations while decreasing the transfer of patients to other centers. This decentralization could also reduce the waitlist for elective cases, while offering to the entire population appropriate and well-timed clinical and surgical care. Moreover, telemedicine and the implementation and constant improvement of neurosurgical training programs could be very helpful to strengthen health care systems in developing countries.[5-7]

The management of patients with multiple intracranial aneurysms represents a greater challenge in a developing country for the above-mentioned reasons. Our patient, other than being female and living at $\geq$3000 m above sea level, had no risk factors for or a family history of intracranial aneurysms. Hence, the discovery of 7 cerebral aneurysms in such a patient is rare. Furthermore, the patient presented mirror posterior communicating artery

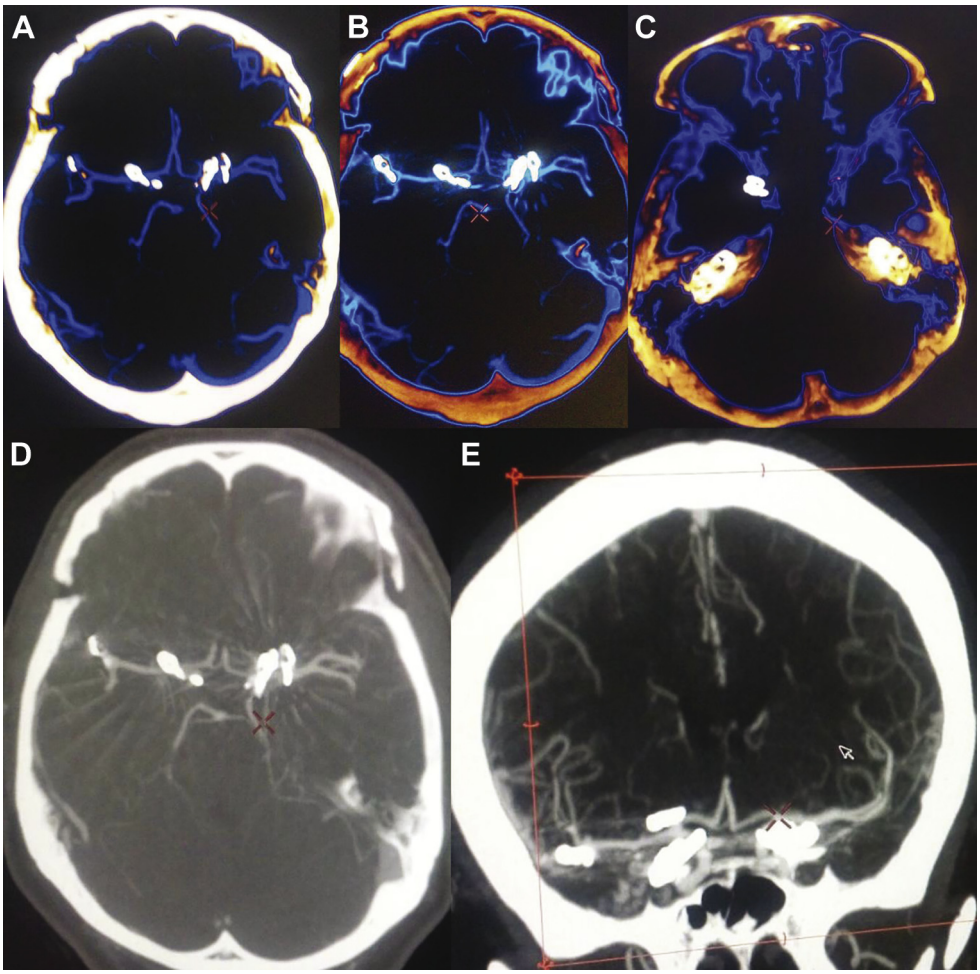

Figure 4. Axial (**A**–**D**) and coronal (**E**) postoperative computed tomography scans revealing complete occlusion of all aneurysms and the patency of the parent vessels without evidence of hemorrhage or ischemia.

aneurysms: the frequency of mirror aneurysms ranges widely from 5% to 40% of all patients with multiple aneurysms.[8-10]

The occurrence of mirror posterior communicating artery aneurysms, together with the fact that the other aneurysms were located in the anterior circulation, suggests a specific trigger in the embryologic prosencephalic region during cephalic segmentation and not before it. The anterior circulation originates from the prosencephalic segment.[8-10] Although the patient had no family history of cerebral aneurysms, genetic counseling as well as screening of family members was recommended.

Single-stage management of 7 cerebral aneurysms is infrequent, and, to the best of our knowledge, this case represents the largest number of aneurysms surgically treated in 1 session.[9,11,12] Strict guidelines for the management of multiple aneurysms are lacking. Endovascular techniques may represent a less invasive treatment modality and may lead to a faster recovery. However, these techniques are extremely expensive and may not be within the budget of health care systems in developing countries. There are only a few neuroendovascular units in Perú, and all are located in the capital district. Moreover, coiling for very small aneurysms is technically challenging and frequently requires adjunctive techniques such as balloon-assisted or stent-assisted coiling, which may require subsequent antiaggregation and anticoagulation for many months after the procedure. Additionally, coiling is associated with higher intraprocedural rupture rates in very small aneurysms compared with larger ones.[4,13,14]

Microsurgical clipping of very small aneurysms is also particularly demanding. Nonetheless, various techniques, such as the double-clip technique, may be effectively employed for their complete occlusion, while preventing clip slippage.[4]

The optimal approach to manage multiple cerebral aneurysms surgically is controversial. According to some authors, a single-stage surgery may increase surgical time and neurovascular manipulation, resulting in worst outcomes. Several reports claim that 2 separate craniotomies in the same sitting for dealing with multiple aneurysms may results in high morbidity

and mortality rates. Hence, a multistaged approach should be preferred.[15-17]

Other authors claim that a single-stage definitive clipping should be attempted if possible. Multistage treatment may increase the risk of rupture of unclipped aneurysms and aggravate the psychological burden for the patient. In addition, a single-stage operation is recommended to decrease the hazards of surgery related to cerebral vasospasm, while facilitating postoperative aggressive hyperdynamic therapy without risk of bleeding from untreated aneurysms. Moreover, hospital stay and relative costs may be increased by a multistage treatment.[15-18]

Unilateral multiple intracranial aneurysms are more suitable for single-stage surgical management. Even if contralateral aneurysms may be successfully clipped through an unilateral craniotomy in selected cases and in experienced hands, more distantly located aneurysms require an additional trephination.[15,16,19] In our case, we reasoned that in a setting of subarachnoid hemorrhage the contralateral aneurysms (in particular, the right anterior choroidal artery and posterior communicating artery aneurysms, partially hidden by the carotid artery and the optic nerve), would have been best and safely managed through an ipsilateral approach.[12,20] We could not use indocyanine green videoangiography for an intraoperative assessment of the occlusion of the aneurysms and the preservation of the blood flow in parent and branching arteries because this technique was not available in the new Neurosurgical Center of Trujillo at the time. We could rely only on an old Doppler and on our intuition and experience.

As corroborated by postoperative studies, an accurate clipping technique allowed the complete exclusion of all the aneurysms from circulation, while preserving the normal flow. Even if this does not prevent regrowth, the wrapping of the fusiform segment of the supplying artery, if possible without incorporating any vessel in the wrap, could be applied to reduce the risk of recurrence. The management of multiple intracranial aneurysms should be tailored on a case-by-case basis, in relation to different factors such as the preoperative clinical status, size, location, and projection of the aneurysms and the experience of the surgeon, with the final goal to ensure the best outcome.[9,12,20]

In this regard, we believe that inside the operation room, teamwork coordination (surgeon, assistant, nurses, anesthesiologist) and judicious application of microsurgical principles (i.e., performing every microneurosurgical procedure in a simple, clean, and fast way, while preserving normal anatomy) play a key role.[21,22] This approach may enable neurosurgical operations to be completed in an efficient and safe manner, optimizing the available resources and the chances of a good outcome for the patient. This takes on utmost importance in resource-challenged environments.[21,22]

CONCLUSIONS

Single-stage surgical treatment of 7 cerebral aneurysms in a 58-year-old woman was successfully and safely performed, despite the fact that the latest equipment was unavailable. The value of good teamwork and application of microsurgical principles must be never forgotten, even in the current technologic era. It is not the equipment and the walls that make the difference, but it is the people carrying out the treatment.

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