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Surgical Removal of Jugular Paragangliomas After Stenting of the Intratemporal Internal Carotid Artery: A Preliminary Report

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Objective: **Jugular paragangliomas with extensive involvement of the internal carotid artery (ICA) represent a true challenge for surgeons, especially in the presence of inadequate collateral circulation through the circle of Willis. The aim of our study is to present a preliminary report of our experience with the surgical removal of three such cases using the stenting of the ICA as the method of choice for protecting and preserving the integrity of the artery.** ***Methods:*** **This retrospective study was conducted at Gruppo Otologico, a private referral center for neurotology and skull base surgery. The subjects of our study are three cases of jugular paragangliomas with extensive involvement of the ICA and inadequate collateral circulation. These cases are the first three cases operated at our center after stenting of the intratemporal portion of the ICA.** ***Results:*** **Complete surgical removal of the tumor, including the part involving the ICA, was achieved in all cases. Over a follow-up period of 22 to 30 months, no complications occurred and the patency of the stented arteries was preserved.** ***Conclusion:*** **Although more follow-up is still needed before establishing the exact long-term outcome of stenting the intratemporal ICA, our preliminary report shows that the stent has facilitated the complete surgical removal of the tumor, preserving the integrity of the ICA.** ***Key Words:*** **Internal carotid artery, stent, jugular paragangliomas.**

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INTRODUCTION

The involvement of the internal carotid artery (ICA) by jugular paragangliomas has always presented a significant challenge to skull base surgeons.

The method of choice for ICA management depends mainly on the degree of involvement of the artery as demonstrated by high-resolution computed tomography (CT), magnetic resonance imaging (MRI), and intraarterial digital subtraction angiography (ANG).[1–3] In the relatively simple cases, removal of the tumor preserving the ICA can be achieved by relatively simple measures like skeletonization, subperiosteal/subadventitial dissection, or displacement. In more extensive cases, however, dissection of the tumor from the ICA can prove to be hazardous to the artery. In such situations, we have successfully resected the tumor and the infiltrated vessel using the technique of permanent carotid occlusion when tolerated by the patient.[3] In cases of intolerance to carotid occlusion, our policy was subtotal removal of the tumor or "wait and scan."

Recently, after the appearance in the literature of papers discussing stenting of the ICA for the management of various pathologies,[4–7] and some few cases of stenting the IA for the management of neoplastic involvement of the artery,[8–11] we have decided to introduce this technique in our practice.

In this article, we present a preliminary report of three cases of jugular paragangliomas involving the ICA; we then discuss the indications and the technique of ICA stenting as well as the impact of the stent on surgical removal of the tumor.

CASE REPORTS

Case No. 1

A 37-year-old female patient presented with a right-sided jugular paraganglioma involving the cervical and temporal segments of the ICA with intracranial–intradural extension into the posterior cranial fossa, class C3Di2 according to Fisch's classification (Fig. 1). The preoperative angiography showed inadequate collateral circulation through the circle of Willis (Fig. 2). An attempt of surgical removal was performed without preoperative precautions to the ICA. During surgery, the tumor was found too adherent to the ICA with a high risk of a major injury to the artery. The operation was discontinued after subtotal removal.

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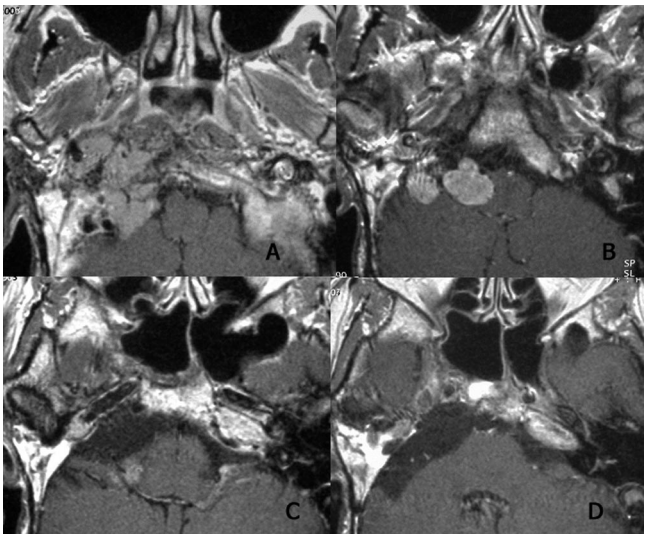

Fig. 1. Axial Gd-enhanced magnetic resonance image showing encasement of the cervical (A) and petrous vertical (B) portions of the internal carotid artery (ICA). (C and D) Extension of the tumor along the horizontal portion of the petrous segment of the ICA.

Three years later, the patient underwent stenting of the ICA. Two Xpert stents, 5 mm in diameter and 40 mm long (Abbott Laboratories Vascular Enterprises, Abbott Park, IL) were inserted in the temporal and cervical segments of the ICA as shown by ANG (Fig. 3) and CT (Fig. 4). Four weeks later, tumor embolization was carried out. Two days later, resection of the tumor involving the ICA was completely achieved. During surgery, the surface of the stent was used as an exact dissection plane between the tumor and the ICA. Furthermore, the presence of the stent facilitated safe and easy manipulation of the ICA. Back and forth displacement of the ICA to dissect 360° of the circumference of the artery was facilitated and the risk of injury to the artery was greatly reduced because of the presence of the stent. The fact that the internal surface of the stent was covered by a layer of neointima abolished bleeding and an almost bloodless field was obtained, thus reducing the total amount of bleeding to a minimal (Fig. 5).

Postoperatively, the patency of the stented ICA was checked out using Doppler ultrasound examination at discharge, 6 and 18 months, and was confirmed at 30 months by MRI.

Case No. 2

A 47-year-old male patient presented with a left jugular paraganglioma encasing the V2 segment of the left vertebral

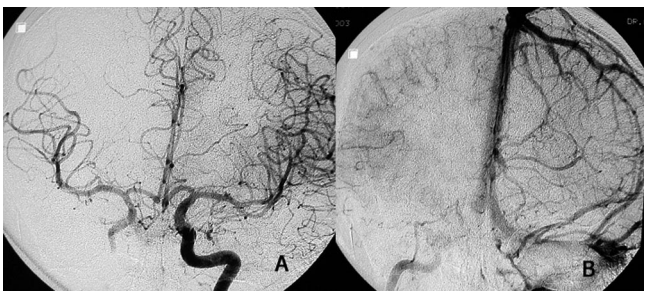

Fig. 2. (A and B) Angiographic evaluation of the anterior portion of the circle of Willis obtained with injection of the left internal carotid artery during manual compression of the right common carotid artery, showing marked asymmetry of the arterial and of the venous phases between right and left hemispheres.

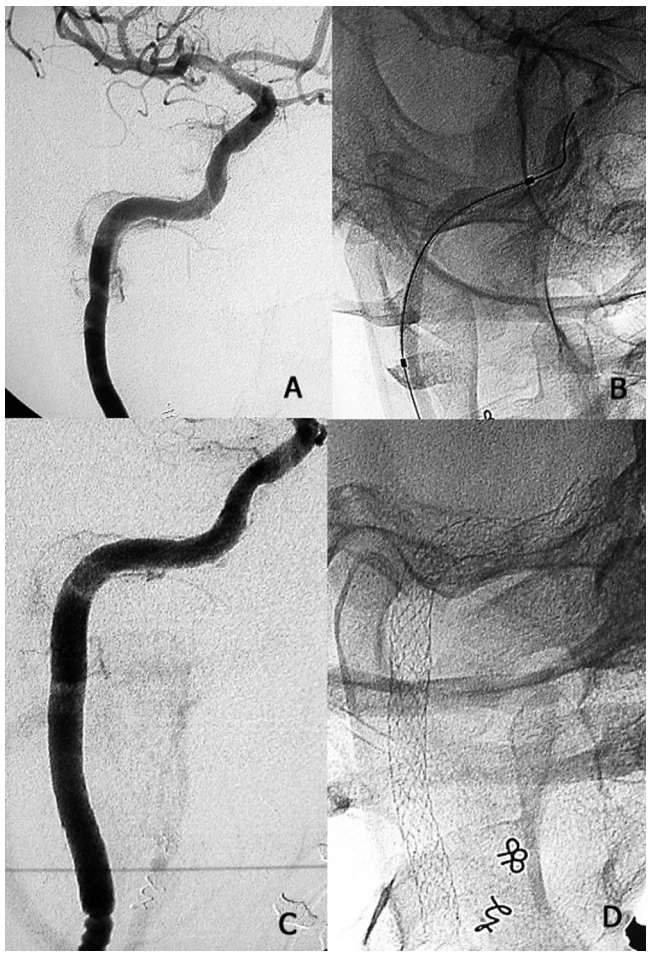

Fig. 3. Right internal carotid artery (ICA) in oblique projection showing extension of the blush along the cervical and petrous segments of the ICA (A) and insertion of the stent (B). Right ICA after stenting of the petrous and cervical portions (C and D).

artery, the distal cervical segment of the ICA, and involving the whole length of the intratemporal ICA, C3Di2 according to Fisch's classification. Preoperative angiography showed inadequate collateral circulation through the circle of Willis.

During the first surgical procedure, we planned a subtotal resection of the tumor, and the pericarotid portion of the tumor was left behind.

After a 5-year period of wait and scan, a progression of tumor growth was shown by MRI. ANG demonstrated a slight stenosis of the vertical portion of the temporal segment of the ICA with vascular supply from the meningohypophyseal trunk (Fig. 6). After the success in case no. 1, the same stenting procedure

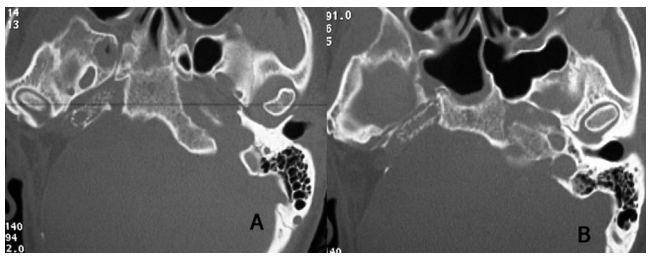

Fig. 4. (A and B) Computed tomography scan of the skull base showing the stent inside the petrous carotid canal.

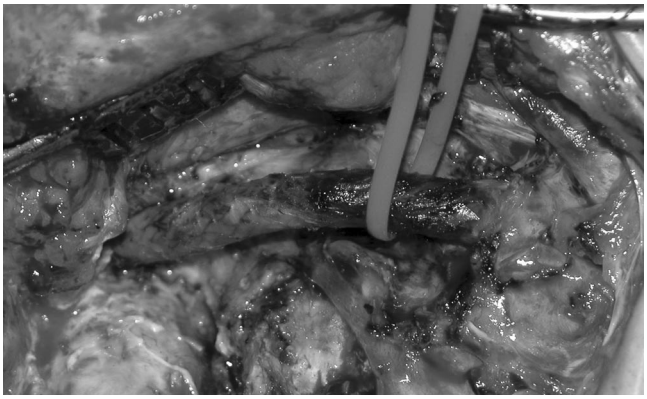

Fig. 5. The view of the internal carotid artery. Tumor removal has been completed. Dissection has been carried out down to the stent in an almost bloodless field.

was used (Fig. 7). Like in the first case, the presence of the stent rendered tumor dissection from the involved ICA exceedingly easy and reduced blood loss significantly.

Doppler ultrasonography 2 years postoperatively confirmed the patency of the stented artery.

Case No. 3

A 42-year-old male patient presented with bilateral jugular paraganglioma. On the right side, both MRI (Fig. 8) and ANG (Fig. 9) demonstrated encasement of the distal cervical and vertical temporal segments of the ICA (C2 De2 according to Fisch's classification).

Initially, we thought it was possible to resect the tumor completely using a simple surgical procedure. The patient was scheduled for preoperative embolization, which was to be followed

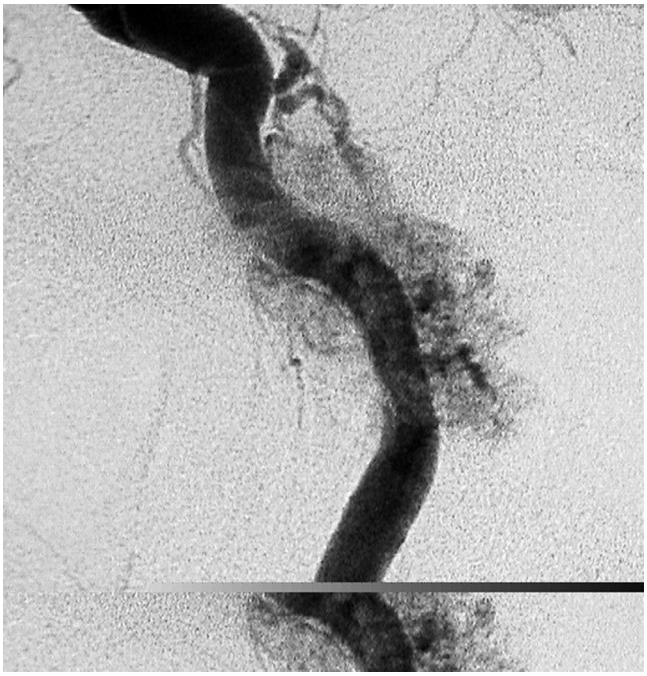

Fig. 6. Digital subtraction angiography in a lateral view showing tumor blush with vascular supply coming from vascular clival branches of the internal carotid artery and stenosis of the distal cervical and vertical petrous portions.

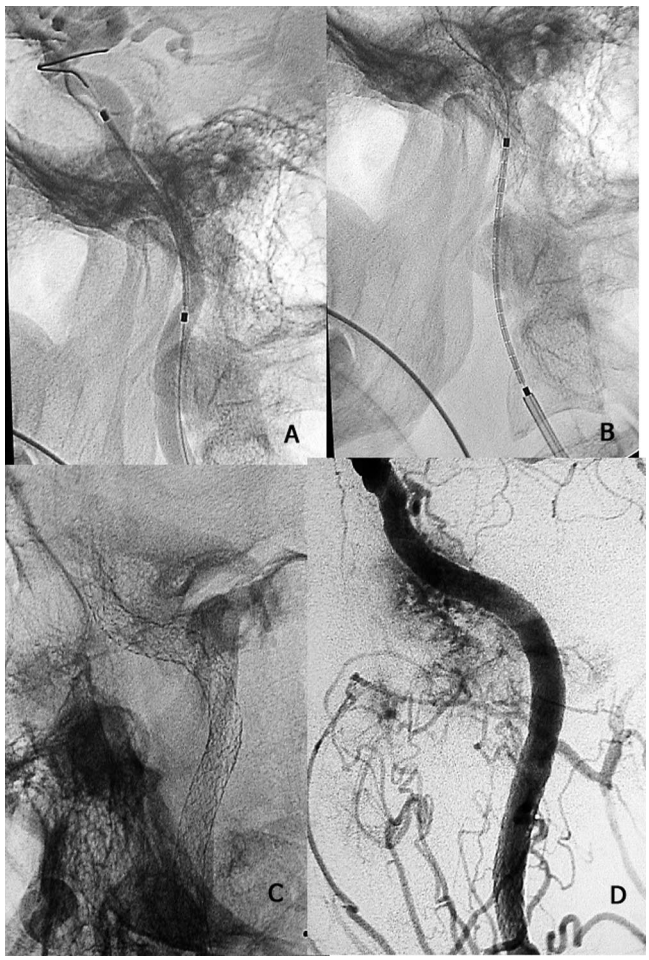

Fig. 7. (A and B) Insertion of the stents in the petrous and cervical portions of the internal carotid artery (ICA). (C) Digital x-ray in oblique projection showing the stents fully deployed in the petrous and cervical segments of the ICA. (D) Digital subtraction angiography of the ICA after stenting showing resolution of the stenosis.

by surgery. By the end of the preoperative embolization, however, the slight stenosis of the distal cervical segment of the ICA (at the entrance to the skull base), which was present before the procedure, was accentuated. This was interpreted as a clear sign of carotid wall infiltration. The planed simple surgical removal was abandoned because of the high risk of injury to the ICA (Fig. 10).

The young age of the patient and the presence of a contralateral jugular paraganglioma were considered to be an indication

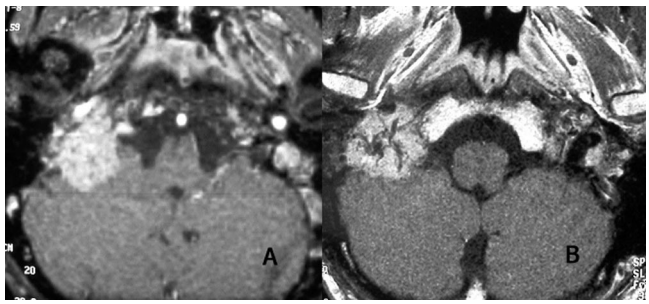

Fig. 8. (A and B) Magnetic resonance image after Gd showing encasement and stenosis of the distal cervical portion of the internal carotid artery.

Fig. 9. Digital subtraction angiography showing tumora blush fed by clival branches and stenosis of the distal cervical portion and vertical petrous portion of the internal carotid artery.

for the preservation of the integrity of the ICA. A single stent procedure was performed followed 2 months later by complete surgical removal of the tumor preserving the patency of the ICA. During surgery, the plane of dissection afforded by the presence of the stent facilitated the dissection of the tumor from the involved segment of the ICA. The absence of bleeding was also noticeable. The patency of the ICA was confirmed by Doppler ultrasonography at 4, 10, and 22 months postoperatively.

DISCUSSION

Extensive involvement of the ICA in jugular paragangliomas is a significant finding with a high risk to the artery; thus, it is influential in the decision-making process. No surgeon can guarantee the safety of the ICA if total surgical removal of such a tumor is to be achieved. In fact, some of the most experienced surgeons have reported intraoperative injury to the ICA with subsequent complications like massive stroke and death.[12,13]

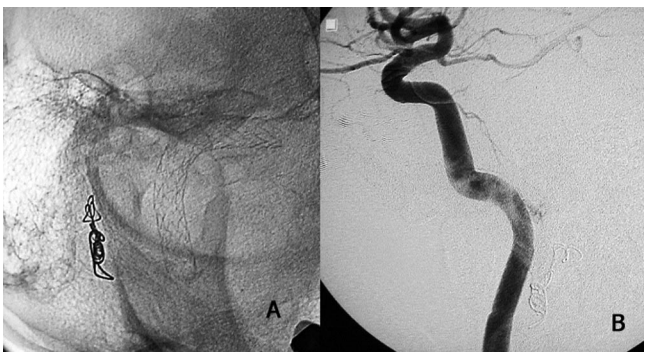

Fig. 10. (A) Digital x-ray film in oblique projection showing the stent in the internal carotid artery (ICA) and coils in the external carotid artery from a previous embolization. (B) Digital subtraction angiography of the ICA in lateral projection showing resolution of the stenosis after stenting.

Consequently, some form of carotid artery management should be used. If preoperative ANG showed the adequacy of the collateral circulation through the circle of Willis, permanent balloon occlusion as described by Fisch is considered to be the simplest and most effective way to avoid intraoperative rupture of the ICA. This procedure is not risk-free, and despite their low rate, these complications can be serious. In addition, this procedure cannot be used in cases of inadequate collateral circulation.

The bypass of the ICA has always been considered the only option in cases of inadequate collateral circulation. Regardless of the technique used, this procedure is a major surgical operation and carries a relatively high risk of restenosis, thromboembolism, and occlusion.

In the literature, there are many reports describing the management of these complications using the endovascular techniques like stenting[4] in addition to few reports on the management of neoplastic involvement of the ICA.[8–11] Such reports were our actual stimulating factor.

In cases in which the tumor engulfs the whole circumference of the ICA, the tumor tissue can be found adherent to the wall of the artery and, in more extensive cases, infiltrating its wall. In such situations, subadventitial dissection is required. This procedure carries a high risk of injury to the ICA, especially in irradiated or previously operated cases. An alternative technique in such complicated cases to enable subadventitial dissection preserving the integrity of the ICA is stenting of the involved segment of the ICA.

In our article, we presented three cases of jugular paragangliomas with various degrees of involvement of the intratemporal segment of the ICA. To enable safe and total surgical excision of these tumors, stenting of the ICA was used as the precautionary measure to avoid its injury.

Before we attempted any of these procedures, we studied the pros and cons of stenting the ICA. To the best of our knowledge, the literature contains only one case of stenting of the intratemporal segment of the ICA for the management of jugular paraganglioma,[8] one case of stenting of the intratemporal segment of the ICA for the management of an endolymphatic sac tumor.[9] In both cases, a covered stent has been used. Moreover, a covered stent was used for the removal of a carotid paraganglioma[10] and a case of stenting the cervical carotid artery for malignant histiocytoma with a bare stent.[11] In all these cases, the surgical removal has been remarkably facilitated as a result of the presence of the stent.

Our knowledge about the complications of TCA stenting is derived from the reports on the endovascular treatment of other lesions, mainly carotid atherosclerosis, trauma, and aneurysms.

When stenting of the ICA is performed to relieve stenosis resulting from atherosclerosis, the most serious acute complication is distal embolization with the risk of definitive stroke. The reason for this complication is the release of emboli from the plaque as a result of the trauma precipitated by the introduction of the stent and not as a result of the stent itself. However, because in paragangli-

omas, the lumen of the artery contains no atherosclerotic plaque, embolization is not a risk, at least early in the process. In their study, Censori et al.[14] have found that carotid stents have a very low emboligenic potential after the early postdeployment period. Other indirect evidences supporting this point of view is derived from the work of Saatci et al.[5] who showed that after applying a covered stent for the exclusion of ICA aneurysms, no mortality or morbidity was seen for a follow-up period of up to 2 years, and from the work of Assadian,[6] in which no occlusion occurred 54 months after stenting the ICA for dissections.

Despite these encouraging studies, the risk of stent thrombosis, however, is still present.[7] Hence, pre- and poststenting antiplatelet therapy should be continued for life. In our practice, patients to be stented are started on 250 mg oral ticlopidine twice a day and 100 mg salicylate daily 1 week before the insertion of the stent. This combination is continued for 1 month after the insertion of the stent. From this point on, oral salicylate is continued for life. The only interruption of this regimen is during the perioperative period. For a duration of 1 week preoperatively and 1 week postoperatively, oral antiplatelet therapy is stopped and low-molecular-weight heparin is administered.

To obtain a safe plan of dissection and to avoid the risk of injuring the ICA at its junction with the tumor, we believe that at least a 5- to 10-mm of tumor-free ICA should be covered by the stent both proximally and distally. To achieve this, it might be necessary to insert two or even three stents. The timing of stent insertion is important. A stent is placed 4 to 6 weeks before the planned date of operation to allow for the formation of a stabilized neointimal lining of the luminal surface of the stent as proved by the histopathologic examination at autopsy by Toma et al.[15] The presence of this neoendothelium, in addition to acting as a support for the stent, has allowed the aggressive tumor resection reaching right down to the stent without the risk of bleeding.

The assessment of the long-term patency of the stented ICA is performed by Doppler ultrasonography. In our three patients, the follow-up period ranged from 22 to 30 months. None of the cases showed signs of stenosis or occlusion of the stented segment of the ICA. In the rare instances in which recurrence of stenosis or thrombosis of the stent occurs, these complications can be managed successfully with angioplasty.

CONCLUSION

We have presented a preliminary report of three examples of jugular paragangliomas with extensive involvement of the ICA in the presence of inadequate collateral circulation. In all cases, complete surgical excision of the tumor involving the ICA and preserving its patency was facilitated by preoperative stenting of the intratemporal ICA. No complications occurred in any of our cases. Although more follow-up is needed before establishing the actual rate of complications, evidence from the literature suggests the long-term safety of ICA stenting.

Although in cases of extensive involvement of the ICA and patent collateral circulation through the circle of Willis, we still use the permanent balloon occlusion. Stenting of the ICA is more convenient than the available alternatives when the collateral circulation is not adequate. The advantages of stenting include avoiding the major surgical operation needed for the bypass operation, preserving the patency of the ICA, facilitating the surgical dissection of the tumor from the artery where the surface of the stent was used as the dissection plan, and combined with preoperative embolization, the neointima formed within the stent provided an almost bloodless field of surgery.

In our opinion, the advantages of stenting the ICA merit further evaluation in all cases of class C3 jugular paragangliomas according to Fisch and in cases in which 360° involvement of any portion of the artery. During the last few months, our senior author has operated additional 12 cases of glomus jugulare tumors using stenting as the precautionary method for the ICA; these cases will be the material of a subsequent study.

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