The emergency use of endografts in the carotid circulation to control hemorrhage in potentially contaminated fields

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We report our experience with the use of endoluminal grafts to control emergency bleeding in two patients with tracheoinnominate fistulas and three patients with carotid blowouts. Systemic infectious complications were not seen. However, rebleeding occurred in one patient, and extensive stent coverage to control bleeding was required in a second. Survival was usually limited by the patient's cancer. There was one long-term survivor without cancer whose tracheostomy was placed for neurologic compromise. A review of the literature for similar cases identified 18 additional endografts placed for carotid blowout and 3 placed for tracheoinnominate fistulas. Overall, infectious complications occurred in only two patients, whereas rebleeding occurred in eight patients. On the basis of these findings, we believe that endografts are useful to control emergency hemorrhage in these two pathologies because treatment is usually palliative, given the poor survival secondary to the underlying disease. However, more extensive graft coverage may be necessary considering the erosive nature of these processes. (J Vasc Surg 2007;46:792-8.)

Tracheoinnominate fistulas and carotid blowouts are two erosive pathologies in the carotid circulation associated with devastating complications. Open repair of either entity has been associated with mortality rates approaching 50%.^{1,2} In addition, the postoperative stroke rate among patients who survive open ligation for carotid blowout has been estimated to be as high as 60%.² In light of these findings, endovascular interventions have been proposed as an alternative to open intervention. The use of covered stents is one such intervention that has been reported, with varying outcomes. Covered stents are an attractive alternative for many reasons. These include the ability to rapidly control hemorrhage from a remote, minimally invasive site while maintaining cerebral flow. However, this also involves the placement of a prosthetic device in a potentially infected or contaminated field. Because of the rarity of these events, there are no large series examining the outcome of this strategy. Therefore, the purposes of this article are to report our experience with the emergency use of endografts in two patients with tracheoinnominate fistulas and three patients with carotid blowouts and to review the literature regarding this treatment strategy. The cases reported entail our institution's total experience with this approach and occurred between September 2003 and July 2006.

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CASE REPORTS

Case 1. A 22-year-old female patient presented with intermittent copious bleeding from her tracheostomy. The tracheostomy was placed in early childhood for severe cerebral palsy, mental retardation, and hydrocephalus. Computed tomographic angiography of the neck and chest suggested tracheoinnominate fistula. Bronchoscopy revealed an erosion of the innominate artery on the anterior wall of the trachea with evidence of recent bleeding. After extensive discussion with the family regarding treatment options, including comfort measures only, a decision for endovascular repair was reached. Repair was achieved by placing an 8×30 mm Wallgraft (Boston Scientific, Natick, Mass) in the innominate and proximal right common carotid artery (CCA) via a 9F sheath after open exposure of the left femoral artery. In addition, a 5F sheath was placed percutaneously in the right femoral artery for simultaneous angiography to guide placement. Completion angiography revealed successful control of bleeding with late filling of the right subclavian artery (Fig 1). After the operation, the patient had no complications and no further bleeding. She was sent home on intravenous antibiotics (clindamycin) for 6 weeks. She was alive without infection at a follow-up of 24 months.

Case 2. A 77-year-old female patient presented with severe bleeding from her tracheostomy. The tracheostomy was placed 2 months earlier after laryngectomy, central node dissection, and radiotherapy for anaplastic thyroid carcinoma. Manual pressure was necessary to control hemorrhage during transport from the referring hospital. Unfortunately, the patient became increasingly unstable and was in extremis upon arrival to the operating room. Expeditious right brachial artery access was obtained by using an open technique, and a 12F sheath was inserted into the artery. Retrograde angiography revealed a bovine arch and diffuse bleeding from the innominate artery, and this partially controlled the hemorrhage. A repeat angiogram showed continued bleeding distal to the graft from the origin of the right CCA. Percutaneous left

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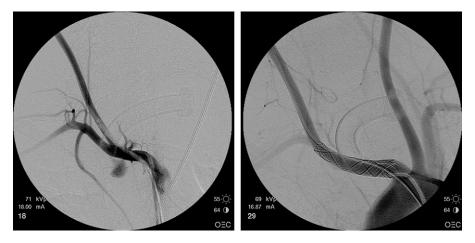


Fig 1. Intraoperative angiography demonstrating extravasation from the innominate artery (*left*) and successful exclusion by an endograft extending into the common carotid artery with late filling of the subclavian artery (*right*).

femoral access and placement of a 12F sheath was performed. The right CCA was selectively cannulated, and a 10×70 mm Wallgraft was then deployed as an extension of the innominate stent into the right CCA. Another 12-mm endograft would have been preferable, but was not readily available at the time. Regardless, there was no type III endoleak, and bleeding ceased from the neck at that point. A 12×20 mm Wallstent (Boston Scientific) was then used to secure fixation of the 10-mm Wallgraft inside the 12-mm Wallgraft. Completion angiogram revealed no endoleak and good flow in the right carotid. An open repair of the left femoral artery, as well as the right brachial artery, was then performed. An axillaryaxillary bypass was also necessary in this case because of severe hypoperfusion to the right upper extremity. The patient sustained a right occipital infarct perioperatively. She was discharged to hospice on postoperative day 7 with a resolving left hemiparesis. She died 1 month later from her malignancy.

Case 3. A 51-year-old male patient presented with bleeding from a recurrent tumor in his left neck. Six months earlier, the patient had undergone total laryngectomy, tracheostomy, bilateral neck dissection, and radiotherapy for laryngeal squamous cell carcinoma. Angiography revealed intense tumor blush fed predominantly by hypertrophied branches of the left external carotid artery (ECA). In addition, there was irregularity of the cervical left internal carotid artery (ICA)-consistent with tumor involvement-as well as adequate cross-filling of the intracranial circulation from the right carotid system. The left ECA and its branches were embolized with polyvinyl alcohol particles ranging from 150 to 250 µm. However, the patient rebled the following day. On the basis of the previous day's angiographic findings, a decision was made to sacrifice the entire left carotid system. A 10F sheath was placed percutaneously in the right femoral artery. An 8 \times 5 cm Viabahn endograft (W.L. Gore & Associates, Flagstaff, Ariz) was deployed in the left CCA and ECA to abruptly occlude flow in the ICA. The CCA and ECA were then embolized with multiple coils. The patient had no postoperative complications and no further bleeding. He was readmitted 1 month later as a result of dysphagia for which he required laparoscopic G-tube placement and lysis of abdominal adhesions. Al-



Fig 2. Exposure of an endograft in the tumor bed secondary to tumor erosion.

though continued tumor erosion led to late exposure of his endograft (Fig 2), he had no systemic infectious complications. He died 4 months later as a sequela of his cancer.

Case 4. A 61-year-old female patient presented with brisk arterial bleeding from her left neck. Five years earlier, the patient had undergone total laryngectomy, radical neck dissection, and radiotherapy for laryngeal carcinoma. In addition, she had a prior failed attempt at esophageal reconstruction via free flap. She arrived in the operating room in extremis with severe acidosis; she was intubated and hypotensive. Angiography revealed bleeding from the proximal left CCA near the base of the wound. An 8×30 mm Wallgraft was deployed in the left CCA from percutaneous right femoral access and seated with a 6-mm balloon, resulting in abrupt control of the bleeding. The femoral artery was then repaired by an open approach. Pectoralis flap coverage of the neck wound was then performed by the head and neck surgeons. However, during that procedure, the patient rebled from distal to the stent. This was controlled by using direct manual pressure

while right femoral access was re-established. Repeat angiography revealed thrombosis of the initial stent and the left CCA. The patient recovered uneventfully but died 4 months later as a result of her cancer.

Case 5. A 78-year-old male patient presented with oropharyngeal bleeding and hemoptysis. The patient was status post total laryngectomy, chemotherapy, and irradiation for laryngeal cancer. Direct laryngoscopy and bronchoscopy were performed, and these revealed arterial bleeding from the anterior wall of the esophagus and necrotic tissue within the esophageal lumen. The esophagus was packed. Angiography revealed a 2-cm pseudoaneurysm of the proximal left CCA with extravasation. A 7 mm \times 5 cm Viabahn was deployed from right femoral access and seated with an 8-mm balloon, and this successfully stopped the bleeding. The patient was discharged on postoperative day 1 but returned 1 week later with a gastrointestinal hemorrhage. Repeat carotid angiography confirmed continued exclusion of the carotid-pharyngeal fistula. The patient died shortly thereafter from multisystem organ failure.

DISCUSSION

This series illustrates some of the challenges inherent to the endovascular management of this process. It is interesting to note that in our small series, the most common difficulty was continued hemorrhage despite apparent initial control of the offending bleeding site. Systemic infectious complications were not seen in our cohort.

To improve our understanding of the expected outcomes of this approach, we reviewed the literature for similar cases. Literature review was performed by using the PUBMED search engine. Bibliographies from all identified cases were cross-referenced for identification of additional cases. Cases were excluded if the endografts were placed for indications other than active bleeding (eg, stable pseudoaneurysm). We identified 18 additional cases of emergency endografts placed in 17 patients for carotid blowout (Table I) and 3 additional emergency endografts placed for tracheoinnominate fistulas (Table II).

Systemic infectious complications were reported in only 2 of the 21 cases of carotid blowout and in none of the 5 cases of tracheoinnominate fistula. Both of the infectious complications occurred in patients with pharyngeal involvement of the carotid blowout. Although the numbers are small, it is reasonable to infer that the higher bacterial load in such cases may have contributed to the infections. One of the infectious complications was a wound cellulitis that resolved with antibiotics. The other was significantly more morbid, consisting of brain abscess from septic emboli. It should be noted that nine additional patients who had carotid blowout with pharyngeal involvement did not develop infectious complications. There were also five patients with delayed transcutaneous exposure of the stent from continued tumor erosion who did not develop systemic infectious complications. Finally, there were only three cases in which the administration of prolonged perioperative antibiotics was documented. None of these cases was associated with infectious complications.

Rebleeding, conversely, was a commonly reported complication in the postoperative course of these patients.

Significant rebleeding was seen in eight patients. This does not include the patient in our series who underwent stent placement because of rebleeding from the CCA after coil embolization of the ECA. In addition, case 2 in our series required additional stent coverage than was expected on the basis of the initial angiogram. Rebleeding has also been recognized as frequent complication after embolization of the carotid for blowout.² This finding should not be surprising given the erosive nature of lesions. In our experience, the exact bleeding site is not always easy to localize, and there is almost certainly a much more diffuse involvement than may be appreciated on angiography. These findings underscore the importance of more generous coverage of landing zones in these cases as compared with more discrete processes such as penetrating trauma. We would recommend covering at least all of the artery that is involved with tumor.

In terms of other complications, stroke was reported in six cases (23%) overall. However, three of the strokes were delayed complications. The periprocedural stroke rate, therefore, was 12%. Two of the periprocedural strokes were thromboembolic, and one was a watershed stroke. All three resolved. The watershed stroke was probably more a result of the tracheoinnominate fistula itself than a technical complication from the endograft placement. The patient was severely hypotensive and hypoxemic for several minutes just before endograft insertion. In addition, digital compression of the innominate artery was being performed, thus further compromising cerebral flow. It should also be noted that two of the three delayed strokes occurred after subsequent coil embolization procedures performed for late recurrent bleeding. The remaining stroke occurred as a result of septic embolism, as previously noted.

Excluding grafts that were intentionally sacrificed by coil embolization, four grafts occluded. One occlusion (case 4 in our series) was believed to be due to the manual compression necessitated by rebleeding in the distal carotid artery. Only one of these four occlusions resulted in stroke. Again, this occurred in the patient with the septic emboli. Postoperative anticoagulation with clopidogrel was documented in seven patients. None of these patients had graft occlusion. However, two of the patients rebled.

In terms of patient survival, most patients with carotid blowout died within months of the event secondary to their cancer. The longest reported survival in this group was 1 year.

In contrast, three of the five patients with tracheoinnominate fistulas have survived with follow-up ranging from 14 to 24 months. Prior studies on survivors of open repair of tracheoinnominate fistulas have shown that longterm survival is uncommon.¹⁵ It is interesting to note that all three of the long-term survivors in this review required tracheostomy for neurologic compromise, as opposed to local pathology. This observation may account both for the improved overall prognosis and for the improved healing at the endograft implantation site. The presence of cancer and prior irradiation are known risk factors for wound complications. Therefore, patients without these risk factors can be expected to have improved results. It should be noted that the only patient in this subgroup who developed recurrent bleeding had metastatic cancer to the trachea and irradiation with an assumed primary tracheoinnominate fistula. Therefore, this patient had a much more aggressive local pathology compared with the long-term survivors in the series.

Comparisons of the results presented in this review with other treatment options is difficult because of the rarity of these cases and the numerous variables that need to be taken into account when outcomes are evaluated. This is especially true for carotid blowout syndrome. For clarification, we excluded cases in which the bleeding source was solely the ECA or one of its branches. In such cases, selective embolization or ligation of the bleeding or parent artery could be performed alone without necessarily compromising cerebral flow. In addition, we tried to include only emergency cases performed for active hemorrhage, as was representative of the five cases at our institution. Unfortunately, most large series include cases of threatened or impending carotid blowout, such as cases of an exposed carotid artery or a carotid artery pseudoaneurysm.

The first reported case of carotid blowout was in 1962 by Borsany.¹⁶ For the most part, standard treatment for the next two to three decades consisted of ligation. Average estimates from several heterogenous earlier studies suggest that both the mortality and major neurologic morbidity rates are somewhere in the 30% to 50% range.¹⁷⁻²² In a rare contemporary series, Upile et al²³ reported results in 11 patients, noting a 91% 1-month survival, two recurrent bleeds, and a 27% incidence of major morbidity. However, the bleeding site was the ECA in six of these patients. The incidence of major morbidity in the remaining five patients (ie, those with CCA or ICA involvement) was 60%.

As with most vascular pathologies, most of the recent literature has focused on endovascular techniques for the treatment of this disease process. The earliest techniques involved sacrifice of the involved artery, specifically by the use of detachable balloons.^{24,25} Perhaps the largest series examining this approach, by Chaloupka et al,²⁶ consisted of 23 procedures in 18 patients, with 2 early deaths, 2 transient ischemic attacks, and no permanent neurologic events. However, only five patients presented with acute carotid blowout, and two of these involved the external carotid system. Of the three patients in their series who would have met our inclusion criteria, all survived, and there was one transient ischemic event. Thus, most of the patients treated with this method were treated on an urgent, rather than emergency, basis. This allowed for test balloon occlusion before permanent vessel sacrifice. It is interesting to note that Chaloupka et al² later reported a series of 12 patients with recurrent carotid blowout after endovascular occlusion, again emphasizing the erosive nature of these lesions. Although it is logical to assume that ablative techniques would decrease the incidence of rebleeding, it is important to recognize that they do not eliminate the problem.

Another important consideration is that the detachable latex balloons used in this study and others are no longer available in the United States. The current alternative, coil embolization, poses an additional inherent risk in that flow is not abruptly ceased during the thrombotic process. Graves et al²⁷ reported a 26% neurologic complication rate in 19 patients undergoing elective coil embolization of the carotid and vertebral arteries with adjuvant proximal balloon occlusion during the placement of the coils. Unfortunately, none of those patients had carotid blowout. Barr et al²⁸ later reported a 16% neurologic complication rate in 19 patients treated with coil embolization, only 6 of which had adjuvant proximal balloon occlusion. This series did include two patients with carotid blowout. It is interesting to note that both cases required detachable balloons for complete arrest of hemorrhage, and one of the patients experienced a fatal stroke. It should be noted that three cases in our series underwent coil embolization for rebleeding after endograft placement. Stroke occurred in two of these

Case 3 represents a novel approach to achieve vessel occlusion with a combination of a stent graft and coils. As such, we believe that it deserves separate consideration. This case was unique in our series in that it was a recurrent bleed now involving the CCA in a patient who recently underwent a full cerebrovascular evaluation. As such, we had the benefit of knowing that the patient's contralateral system was dominant. Our technique produced abrupt occlusion of the ICA, potentially decreasing the risk of stroke during embolization of the CCA and ECA. Another reason that we chose that particular approach is that the current endografts are not tapered, and we were concerned about infolding of the graft or rupture of the ICA from the oversizing that would have been necessitated if the endograft were extended into that vessel.

The data on tracheoinnominate fistulas seem more straightforward. First described in 1879 by Korte,²⁹ tracheoinnominate fistula remains one of the more formidable challenges in surgery. Gelman et al¹⁵ published what many consider the seminal review of this entity, compiling 71 patients from a review of the literature from 1962 to 1994. The authors found that only 40 (56%) of these patients survived more than 2 months. Survival was much greater in the patients undergoing ligation (71%) than in patients undergoing revascularization (16%). The most frequent cause of death in the latter group was recurrent hemorrhage from suture line failure or infectious complications. Case reports of reconstruction by using cleaner inflow or targets (aorto-carotid, aorto-axillary, or extra-anatomic bypass) suggest fewer infectious complications and rebleeding with these techniques.^{30,31} Others have advocated the use of muscle flaps or autogenous conduits.³²⁻³⁴ In terms of alternative endovascular interventions, there has been one report of embolization of tracheoinnominate fistula.35 The case involved a 9-year-old patient with tracheostomy for encephalopathy. The patient was alive with no further bleeding episodes at 1 year of follow-up. Finally, Sessa et al³⁶ reported the use of a stent graft as a temporizing

Study	Clinical history	Bleeding site	Stent	Anticoagulation	Extended postoperative antibiotics	Outcome
MacDonald et al ³	61-y-old man with tongue CA, s/p ND and XRT	Transcutaneous from R CCA	Jostent	NS	Yes (10 days)	Alive at 5 mo
Kwok et al ⁴	63-y-old man with oropharyngeal CA, s/p ND and XRT	Transpharyngeal from R CCA	Jostent	NS	NS	Rebled during surgery, requiring coil embolization; alive at 2 mo
Warren et al ⁵	67-y-old man with multiple oropharyngeal CA, s/p ND and XRT	Pharyngocutaneous from L CCA	Jostent	NS	NS	Rebled at 3 wk, requiring coil embolization complicated by CVA; subsequent exposure of stent; died several months later
	Same patient	Pharyngocutaneous from R CCA	Jostent	NS	NS	Rebled several months later; died before intervention
	44-y-old woman with multiple CA, s/p ND, XRT, and tracheostomy	Transpharyngeal from origin R ECA	Jostent	NS	At 1 mo	Cellulitis 1 mo after surgery; subsequent stent exposure; died at 4 mo
Bates and Shamsham ⁶	42-y-old man with squamous cell CA of soft palate and uvula	Transcutaneous from R ECA	Wallgraft	Clopidogrel 1 mo	NS	Alive at 6 mo
Lesley et al ⁷		ICA	PTFE- covered Palmaz	ASA/clopidogrel	NS	Alive at 3 mo
	46-y-old man with squamous cell CA, s/p ND	ICA	Wallgraft	ASA/clopidogrel	NS	Alive at 3 mo
	41-y-old man with squamous cell CA, s/p ND and tracheostomy	Origin ECA	Wallgraft	ASA/clopidogrel	NS	Alive at 1 y
Simental et al ⁸	69-y-old woman with unspecified CA, s/p XRT	Transcutaneous from L CCA	Wallgraft	NS	NS	Exposed stent at 3 wk; rebled at 6 wk, requiring coil embolization complicated by CVA and death
	47-y-old man with lingual CA	Transcutaneous from L CCA	Wallgraft	NS	NS	Gradual exposure of stent with thrombosis; died at 2 mo
Desuter et al ⁹	Squamous cell CA of pyriform sinus, s/p ND and XRT	Transpharyngeal from ICA	Jostent	Abciximab after CVA	NS	Perioperative stroke, resolved after treatment with abciximab; died at 7 mo
	Squamous cell CA of pyriform sinus, s/p XRT and tracheostomy	Transpharyngeal from bulb	Fluency	Heparin/urokinase after CVA	NS	Perioperative stroke, resolved after treatment with heparin and urokinase; alive at 5 mo
Kim et al ¹⁰	62-y-old man with esophageal CA	Transcutaneous from CCA	Niti-S	Clopidogrel	NS	Rebled at 11 d; alive at 2 mo
	57-y-old man with nasopharyngeal CA	Transpharyngeal from bulb	Niti-S	Clopidogrel	NS	Rebled at 6 wk; died at 5 mo
	68-y-old man with laryngeal CA	Transpharyngeal from CCA	Niti-S	Clopidogrel	NS	Alive at 1 d
	61-y-old man with hypopharyngeal CA	Transpharyngeal from bulb	Niti-S	Clopidogrel	NS	Alive at 1 d

Table I. Endoluminal graft placement for carotid blowout

Wallgraft (Boston Scientific, Natick, Mass); Viabahn (W.L. Gore & Assoc., Flagstaff, Ariz); Jostent (Jomed Corporation, Rancho Cordova, Calif); Fluency (Bard Peripheral Vascular, Tempe, Ariz); Niti-S (Taewoong Medical, Seoul, Korea); Passager (Meadox, Oakland, Calif).

Study	Clinical history	Bleeding site	Stent	Anticoagulation	Extended postoperative antibiotics	Outcome
Chang et al ¹¹	52-y-old man with squamous cell CA of hypopharynx, s/p XRT	Transpharyngeal from L bulb	Wallgraft	NS	None	Exposed, thrombosed stent in hypopharynx leading to septic emboli and brain abscess with CVA, alive at 6 mo
This study	51-y-old man with laryngeal CA, s/p ND, XRT, and tracheostomy	Transcutaneous from L CCA/ ECA	Viabahn	None	None	Intentionally coiled during surgery; subsequently exposed stent; died at 4 mo
	61-y-old woman with laryngeal CA, s/p ND, XRT, and laryngectomy	Transcutaneous from L CCA	Wallgraft	None	None	Rebled during surgery, thrombosed with manual compression; died at 4 mo
	78-y-old man with laryngeal CA, s/p ND, XRT, and laryngectomy	Transcutaneous from L CCA	Viabahn	ASA	None	Gastrointestinal bleed and death at 1 wk

CA, Cancer; s/p, status post; ND, neck dissection; XRT, irradiation; R, right; CCA, common carotid artery; NS, not stated; L, left; CVA, cerebrovascular accident; ECA, external carotid artery; ICA, internal carotid artery; PTFE, polytetrafluoroethylene; ASA, aspirin.

Table II.	Endoluminal	graft plac	cement for	tracheoin	nominate	fistula

Study	Clinical history	Stent	Extended postoperative antibiotics	Outcome
Deguchi et al ¹²	37-y-old man with dysphagia after resection and XRT of medulloblastoma of the cerebellum 14 y before	Passager	Cefazolin/piperacillin for 4 wk	Alive at 14 mo of follow-up without infection
Wall et al ¹³	47-y-old woman with metastatic adenoid cystic CA to trachea/ lungs treated with XRT	Viabahn	NS	Died at 6 wk after open conversion for bloody tracheal secretions and erosion of stent into trachea
Vianello et al ¹⁴	16-y-old man with Duchenne muscular dystrophy	Wallgraft	NS	Alive at 14 mo of follow-up without infection
This study	22-y-old woman with severe cerebral palsy	Wallgraft	Clindamycin for 6 wk	Alive at 24 mo of follow-up without infection
	77-y-old woman with anaplastic thyroid CA, s/p , ND, XRT, and tracheostomy	Wallgraft	No	Perioperative watershed CVA; died at 4 wk

XRT, Irradiation; s/p, status post; CA, cancer; NS, not stated; ND, neck dissection; CVA, cerebrovascular accident.

measure followed by revascularization with a cryopreserved allograft 12 hours later. The case involved a patient who developed hemoptysis after tracheal biopsy secondary to puncture of the innominate artery by a tracheal ring. It is interesting to note that all bacteriologic test results at the time of the second operation (ie, explantation and open repair) were negative.

In summary, on the basis of our findings, the use of endovascular stents to control hemorrhage from carotid blowout and tracheoinnominate fistula compares favorably to other reported modalities of treatment for these disease processes. Considering the nature of these lesions, infectious complications are relatively infrequent but do occur. Prolonged antibiotics may be of benefit, especially when there is pharyngeal involvement. Recurrent bleeding episodes, conversely, are common, likely because of the erosive nature of these lesions, and extended coverage zones should be considered.

Long-term survival in patients with cancer is severely limited. Therefore, these procedures should be considered palliative. In that context, the short-term benefits of this approach seem to warrant its use. In patients with tracheoinnominate fistula after prolonged tracheostomy for neurologic compromise, endografts seem to be durable. However, the world experience with this approach is small.

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