CASE REPORT

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CERVICAL HEMORRHAGE DUE TO SPONTANEOUS RUPTURE OF THE SUPERIOR THYROID ARTERY: CASE REPORT AND REVIEW OF THE LITERATURE

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Abstract: *Background.* Beneath the different reasons for cervical masses, a spontaneous hemorrhage presents a rare and life-threatening condition.

Methods and Results. We present the rare case of a 62-year-old man who was presented with a dramatically enlarging cervical mass causing respiratory distress because of upper airway compression. An endotracheal intubation was lifesaving and avoided tracheotomy. A CT scan revealed a hematoma in the region of the left external carotid artery. An emergency angiography embolized a ruptured branch of the superior thyroid artery and surgery evacuated the hematoma. We discuss the rarity of the condition, reasons for a spontaneous rupture of the artery, and the diagnostic and treatment strategy. In addition, we review the literature on spontaneous thyroid artery hemorrhages, which, up to now, have been described only for the inferior thyroid artery.

Conclusion. We conclude that the optimal management for cases of cervical hematoma is intubation, diagnosis, and angiography before surgery. © 2009 Wiley Periodicals, Inc. *Head Neck* **32:** 1277–1281, 2010

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The differential diagnoses of cervical masses are versatile and include benign and malignant neoplasms, inflammatory lesions, and hematoma. Beneath this, a spontaneous hemorrhage presents a rare and life-threatening condition due to potential cerebral hypoxia and airway compromise.[1] It results in cervical hematoma and is characterized by tracheal, esophageal, and vascular compression, tracheal displacement, and subsequent endolaryngeal and subcutaneous bruises.[2] The etiologies of spontaneous cervical hemorrhage may be diverse, comprising ruptured arteries, ruptured arterial aneurysms, and artery dissection. The clinical course has a 2-step pattern. The initial limited extravasation is followed by sudden enlargement and ends with suffocation and eventually death.[3]

To the best of our knowledge, we present the first report of a case of spontaneous rupture of the superior thyroid artery (STA). We discuss diagnostic and therapeutic procedures and give an overview on hemorrhages caused by the

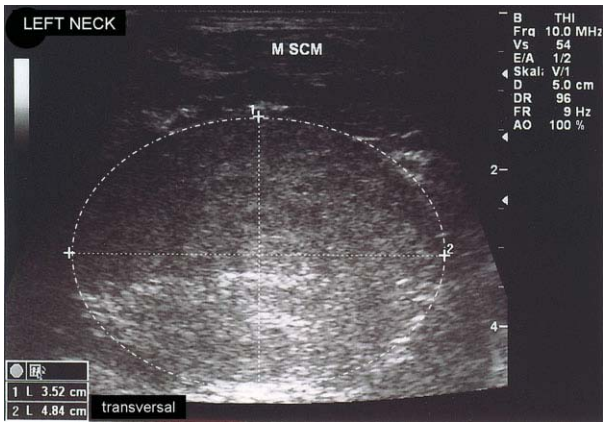

FIGURE 1. Ultrasound image shows the dimensions and the structure of the cervical mass M. SCM, sternocleidomastoid muscle. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

inferior thyroid artery, which seem to be more common.

CASE REPORT

A 62-year-old man initially presented to our outpatient department with a left-sided neck swelling, which suddenly began with increased strain of the skin 4 hours before admission. The patient had no stridor but complained of dysphagia. He denied symptoms of an upper respiratory tract infection, a history of trauma, past operations, ingestion of any foreign objects, a bleeding diathesis, or other medical problems. Clinical examination revealed a large, firm, painful, and nonfluctuant swelling on the left side of the neck extending from the mandible to the clavicle. Indirect hypopharyngoscopy showed an edema of the left aryepiglottic fold. His medical history was empty, remaining ear, nose, and throat (ENT) examination was without pathologic findings, and standard laboratory parameters were all within normal range.

Immediate ultrasound scan showed a huge, inhomogeneous mass adjacent to the left thyroid lobe with transversal diameters of 4.8 $\times$ 3.5 cm (Figure 1). At this time, the first working diagnosis was a thyroid gland carcinoma or neck malignancy. Nevertheless, on repeatedly questioning, the patient assured us that the swelling was new. Therefore, and because of increasing size of the swelling with beginning dyspnea, a native CT and a contrast-enhanced angiographic CT were rapidly performed. The CT scans showed a hematoma-like lesion within the left neck measuring 10 $\times$ 8 $\times$ 6 cm with distinct dislocation of the trachea to the right side (Figure 2). The thyroid gland seemed normal in size and shape with only a slight compression of the left lobe. Meanwhile, the symptoms of dyspnea and dysphagia worsened and the swelling enlarged. For airway protection, an orotracheal intubation was performed without problems and, in suspicion of an acute cervical hemorrhage, an angiography was carried out with the patient under general anesthesia. Entering the right common femoral artery, the left STA was visualized being stretched and compressed. In the direct vicinity, a slight extravasation of contrast medium was apparent. The left STA was selectively catheterized, and bleeding coming from the final branches of the STA could be seen (Figure 3A). The decision was made to embolize the final arterial branch of the STA. Multiple mechanical detachable microcoils with diameters between 2 and 3 mm were positioned

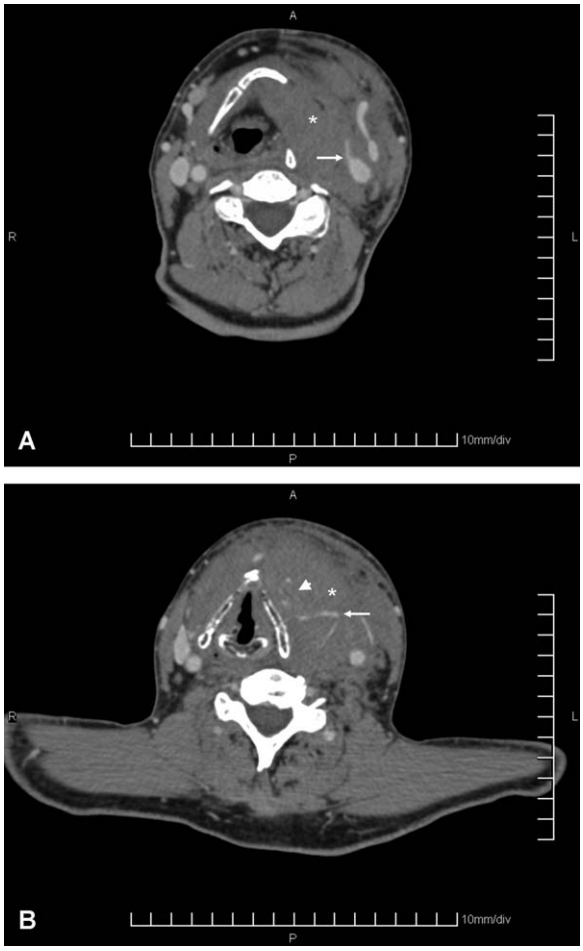

FIGURE 2. Contrast-enhanced angiographic CT scans shows the dimensions of the cervical hematoma (*) with lateralization of the trachea. Departure of the left superior thyroid artery from the external carotid artery in height of the bifurcation **(A, arrow)** and branching of the superior thyroid artery **(B, arrow)** with extravasation of contrast media within the hematoma (arrowhead).

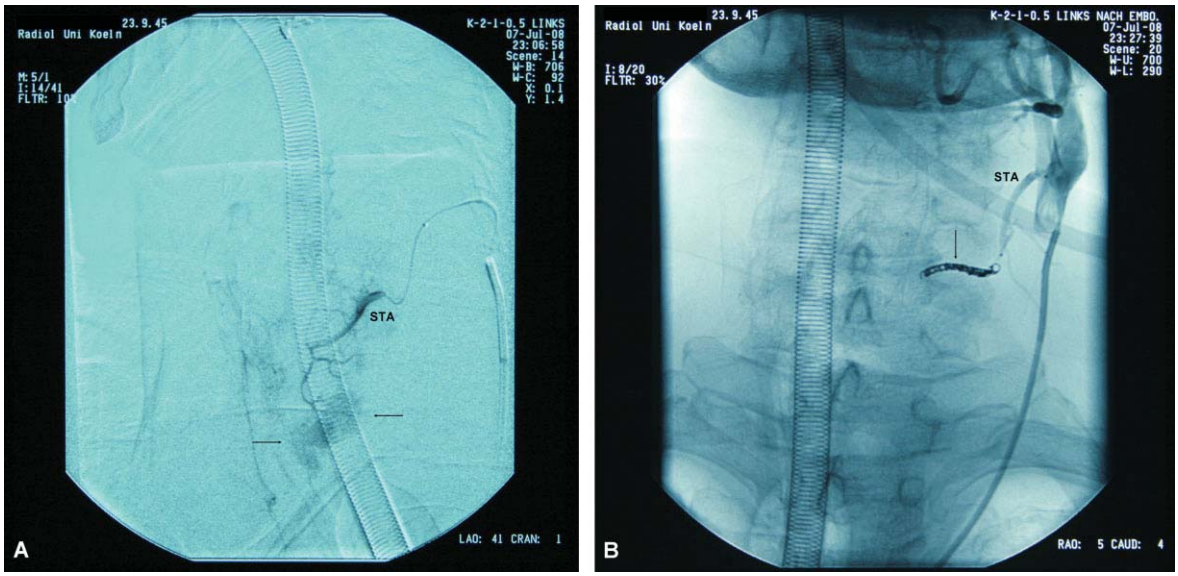

FIGURE 3. **(A, arrows)** Angiogram shows the bleeding coming from the final branches of the superior thyroid artery (STA). **(B)** After embolization, the vessel was completely closed in height of the coils (arrow) and no more extravasation of contrast solution was seen. [Color figure can be viewed in the online issue, which is available at wileyonlinelibrary.com.]

intraluminally. In the closing control, the vessel was completely obstructed in height of the coils and no extravasation of contrast media was detectable anymore (Figure 3B). By displaying the left subclavian and the right common carotid artery, other leakages were ruled out. Furthermore, there was no evidence of aneurysms or arterial dissections.

After angiography, the patient underwent surgical exploration of the left neck. A large organized hematoma in the parapharyngeal space was found and evacuated. The hematoma masses were in the direct vicinity of the thyroid cartilage, the sternocleidomastoid, and the prelaryngeal muscles reaching caudally into the upper mediastinum. The thyroid gland was without macroscopic pathologic findings. After insertion of a drain, the patient was transferred to the intensive care unit postoperatively for 1 night for observation. No tracheotomy was needed.

The following day, after immediate extubation, recovery was uneventful. The patient was discharged 7 days after the operation without any signs of dyspnea or dysphagia. The endolaryngeal edema was gone. Six months later, the patient was in good overall condition and no more bleeding events had occurred.

DISCUSSION

Without a history of trauma, the patient developed a left parapharyngeal hematoma. At first examination, he was presented with the clinical triad of tracheal and esophageal compression, lateral tracheal displacement, and subsequent swelling in the lateral neck. This triad of symptoms associated with cervicomediastinal hematoma had already been reported by Capps in 1934.[4] The rarity of this entity was the reason for a wrong working diagnosis in our case. Only because of the progressive swelling and dyspnea, the patient underwent emergency diagnostic workup and subsequent life-saving therapy.

Besides iatrogenic, traumatic, or tumor-associated etiologies, the spontaneous bleeding from a thyroid artery is a rare incident and in the majority of cases it is caused by a ruptured aneurysm. Aneurysms of the branches of the supra-aortal arteries are extremely rare. They are mainly derived by trauma, atherosclerosis, or central venous cannulation.[5] There exist no reports on spontaneous STA hemorrhage. However, our literature review resulted in 12 cases of spontaneous bleeding from the inferior thyroid artery in the past 50 years (Table 1). In only 1 patient was the etiology inexplicable, whereas in 10 cases an aneurysm was found as the cause (in 1 case the etiology was not stated). Of the resected specimens, 2 were found to be primary dissected aneurysms of the inferior thyroid artery. All others were degenerative aneurysms involving the full thickness of the arterial wall. Atherosclerotic origin was present in the majority of cases, even though media necrosis and posttraumatic false aneurysms can occur. In some cases,

Table 1. Literature review of spontaneous inferior thyroid artery haemorrhage.

Reference	Year	Side	Age, y	Sex	Etiology	Presentation	Treatment	Complications	Outcome
Doumanain[6]	1959	Right	60	Male	Aneurysm	Hoarseness	Surgical excision	None	Survival
Golby[7]	1965	Left	65	Male	Dissected aneurysm	Respiratory distress	Tracheotomy, surgical excision	None	Survival
Martin[8]	1974	Left	43	Female	Aneurysm	Respiratory failure	Surgical excision	None	Survival
Habib[9]	1977	Left	67	Female	Aneurysm	Neck swelling, Respiratory distress, dysphagia, hoarseness	Tracheotomy, observation	Erosion of the tracheal wall with bleeding into the trachea	Death
Lin[10]	1978	Right	80	Female	Dissected aneurysm	Respiratory failure/ dyspnea	Observation	None	Death
Beal[11]	1987	Left	78	Male	Aneurysm	Respiratory arrest	Embolization	None	Survival
Watson[12]	1994	Right	62	Female	Aneurysm	Dysphagia, hoarseness	Embolization, ligation, surgical excision	None	Survival
Kos[13]	2001	Not stated	72	Male	Not stated	Cervical and thoracic pain	Gelfoam plug embolization	Hemothorax	Survival
Heckenkamp[14]	2003	Right	76	Female	Aneurysm	Respiratory failure, dysphagia, hematoma	Intubation, coil embolization	None	Survival
Terzi[15]	2004	Left	53	Female	Aneurysm	Dyspnea, neck swelling, pain	Embolization	None	Survival
Garrett[16]	2005	Left	44	Female	Aneurysm	Pain, neck swelling, respiratory distress	Intubation, tube thoracostomy, coil embolization, transfusion	Hemothorax	Survival
Bageacu[17]	2005	Right	44	Female	Idiopathic	Pain	Intubation, tracheotomy, surgical exploration, transfusion	Mediastinal hematoma, pleural effusion	Survival

coughing or sudden physical work were described as preceding events. Main complaints were respiratory distress, hoarseness, dysphagia, pain, and cervical swelling. If a diagnosis was made by surgical exploration or angiography, all patients survived. The 2 deaths occurred either before a diagnosis could be established or without the benefit of a diagnostic workup.

We, for the first time, report on a patient who suffered from a spontaneous hemorrhage arising from the STA. According to the literature, a ruptured aneurysm could be possible, as ruptured aneurysms need not necessarily be seen during angiography. In our case, the ruptured artery did not show any pathologic findings, either macroscopically or radiologically. No signs of thyroid gland abnormalities were seen during diagnostic imaging or surgery as well. It is known that atherosclerosis might lead to fragile vessels. Major risk factors include age, hypertension, diabetes, hypercholesterolemia, and smoking habits. Interestingly, our patient suffered from none of these, and no predisposing vessel abnormalities, aneurysms, or dissections were seen. Therefore, idiopathic rupture of the STA must be assumed in this case.

What we consider extremely important is that complex diagnostic procedures should be undertaken only after securing the airway. Risk of suffocation by airway compromise is real and has been documented by other authors.[18] This may occur at any moment in the clinical course. We believe that the importance of securing the airway as early as possible cannot be overestimated and that the diagnostic and treatment procedures should only be undertaken after this has been done. Otherwise airway protection may get extremely difficult and needs to be secured by means of fiberoptic intubation or tracheotomy, or it may even become impossible. Another issue that we would like to emphasize is the interventional and surgical procedure. During angiography, the bleeding vessel was detected and could be embolized in 1 step accomplishing definite hemostasis. Therefore, angiography remains a highly important diagnostic procedure and, if possible, embolization seems to be a safe and feasible alternative to open surgery of the ruptured vessel. Nevertheless, surgery is essential to evacuate the hematoma and thus accelerate the extubation of the patient. During surgery, the incision must be wide enough to allow exploration of the whole hematoma by mobilization of the neighboring structures.

CONCLUSION

The rare incident of bleeding from a ruptured vessel has to be kept in mind as a differential diagnosis of an acute neck swelling. Cervical hemorrhage should optimally be managed in the following order: intubation, diagnosis, and therapy by intervention and/or surgery.

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