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Case Report

50-Year history of congenital hemoptysis: A case of gastro-duodenal oral choristoma



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ABSTRACT

A case of life-long hemoptysis in a 50-year-old woman found to be caused by a bleeding gastro-duodenal oral choristoma is presented. The rare nature of the condition could have led to it being misdiagnosed. She had been managed as a case with more common causes of hemoptysis. The clinical value of lessons learned from this case lies in the infinite nature of possible causes of symptoms and signs observed in patients; every case should be treated as unique, or potentially so. The step-by-step scientific method without undue bias in favor of the use of inexhaustive checklists, of causes of symptoms and signs, works best for management of every patient, as all cases are unique. Causes of hemorrhage can hardly be exhaustively listed.

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1. Introduction

There are many differential diagnoses for hemoptysis, which may be caused by lesions related to the aero-digestive tract. These lesions are most commonly bronchitis [1], pneumonia [2], lung cancer and tuberculosis [3]. Gastro-duodenal oral choristoma has not been found to be listed in the literature as one of the possible causes of hemoptysis. We present a rare case of previously intractable congenital hemoptysis caused by gastro-duodenal oral choristoma.

2. Case report

A 50-year-old woman was supported to the emergency room with a complaint of copious bleeding from the mouth. She collapsed immediately on arrival and was resuscitated. History revealed she had lost an unusually large amount of blood from her oral cavity over the previous 24 h. She was a known case of congenital hemoptysis associated with intermittent left submandibular swelling, and

had continually spat blood-stained saliva. It was thought in 1969 to be a hemangioma in the submandibular region because it produced copious amount of blood intraorally when massaged; the lesion was left alone as attempted surgery was abandoned. Other clinicians had treated the swelling as a chronic lymphadenitis resulting from periodontitis, for which she had had several dental extractions. Blood discharge was reported to be from around the periodontium. She subsequently became a known hypertensive (largely controlled) at 45 years. Intraorally, a careful search revealed the source of bleeding to be from a point below the laterally spread tongue, where teeth had been extracted, as shown in Fig. 1. Teeth present were the third molar in the left upper and lower quadrants, and all molars and premolars in the right upper and lower quadrants. All other teeth had been extracted. All teeth present showed signs of attrition, erosion and abrasion. Investigations revealed that her Hb was 9.1 mg/dL. Incisional biopsy of tissue of the bleeding point revealed it to be a gastro-duodenal oral choristoma (Fig. 2). T2 weighted MRI scan showed multifocal lobulated hyperintense masses in the left submandibular region, as shown in Figs. 3–6.

Excision of lesion was carried out, as shown in Fig. 7, under general anesthesia and patient recovered uneventfully. She has been followed up for 7 years without any complication or episode of hemoptysis, as shown in Fig. 8. Tooth-wear and loss also ceased.

* Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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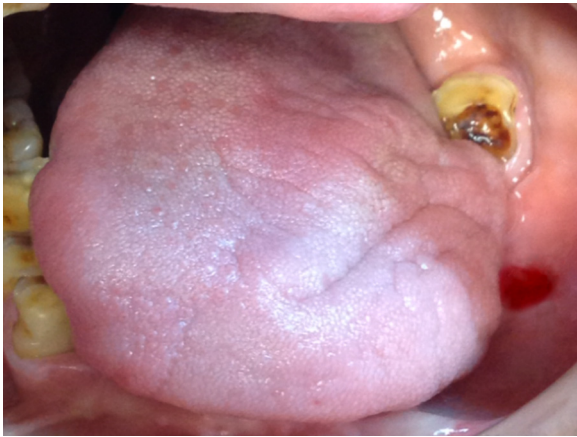


Fig. 1. Intraoral view: blood discharge seen anterior to third molar in lower left quadrant.

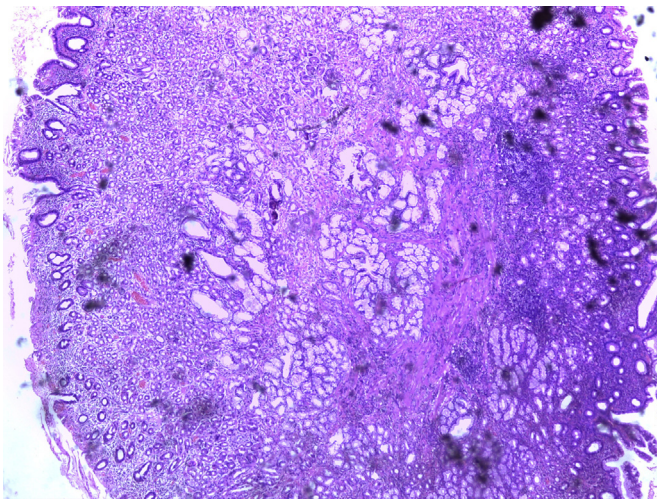


Fig. 2. Photomicrograph of incision biopsy of choristoma showing gastro-duodenal tissue. Stain: Hematoxylin and Eosin. Magnification: 10 \times .

3. Discussion

It is very unusual for hemoptysis to be observed at birth. The patient in this report suffered the condition from birth until she

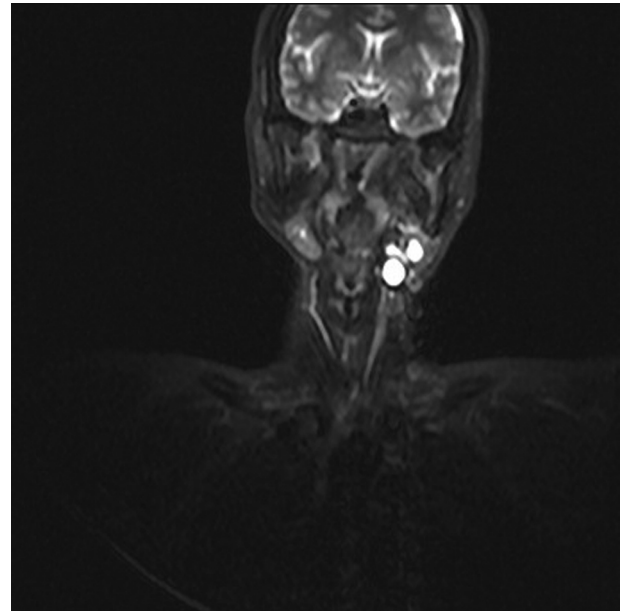


Fig. 4. Coronal view: medio-lateral extent of choristoma shown.

was 50 years old. We have not found any literature that lists gastro-duodenal oral choristoma as a cause of hemoptysis. The clinical value of lessons learned from this case lies in the infinite nature of possible causes of symptoms and signs observed in patients; every case should be treated as unique. The step-by-step scientific method, employed without bias, works best for management of every patient, as all cases are unique. Actual cause of observed effects must be ascertained; this is why clinicians are scientists. We must obtain adequate evidence before any surgical or other action is embarked upon. The first attempt at surgically treating this case would have been assured of success if angiography available in the hospital at that time had been done to eliminate the possibility of the lesion being associated with large blood vessels. When such a case presents, the predilection for many clinicians is to run through a checklist of known causes of hemoptysis starting with the commonest and eliminating them until the clinician is stuck with one or a few. Often, treatment and investigation are carried out based on that one or few left on the checklist.

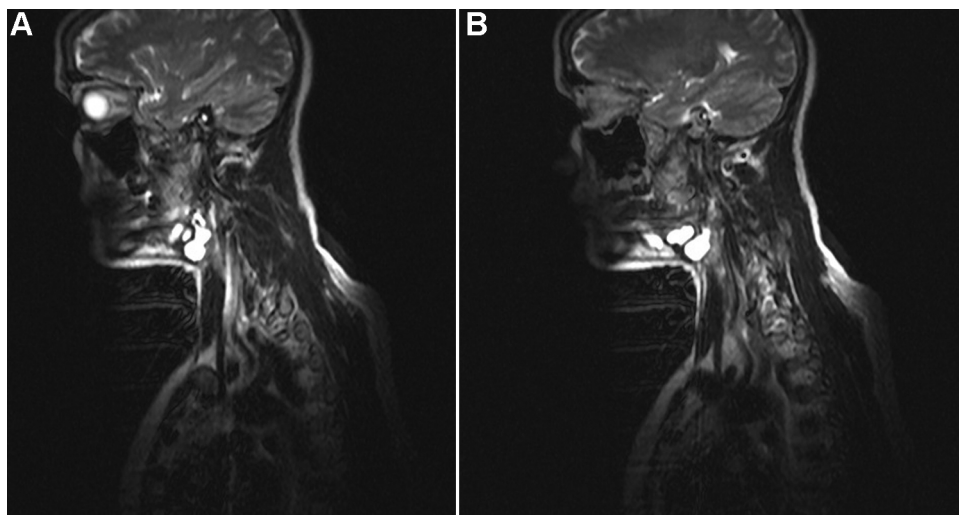


Fig. 3. T2 weighted MRI scan: sagittal view showing multifocal lobulated hyperintense masses in the left submandibular region.

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