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Case report

Endoscopic intranasal control of hemorrhage from unexpected ethmoidal hemangioma during conchoplasty: A case report

Brihaspati Sigdel [a], Dipendra Kandel [a], Mitesh Karn [b,*]

[a] Department of Otolaryngology, Gandaki Medical College Teaching Hospital and Research Center, Pokhara, Nepal
[b] School of Medicine, Gandaki Medical College Teaching Hospital and Research Center, Pokhara, Nepal

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ABSTRACT

Introduction and importance: Hemangiomas of paranasal sinuses are rare. Unexpected hemangiomas of this region can bleed profusely leading to operative morbidity and mortality. Hemangiomas of paranasal sinuses may be associated with concha bullosa.

Case presentation: We report a case of 41-year-old female who presented with difficulty in nose breathing and headache since two years. She was diagnosed with concha bullosa with deviated nasal septum and was planned for endoscopic septoplasty and conchoplasty. During the procedure, profuse, unexpected hemorrhage resulted from the undiagnosed hemangioma which was managed by endoscopic intranasal cauterization of anterior ethmoidal artery.

Discussion: Hemangiomas of paranasal sinuses such as ethmoidal sinus are rare. These are slow growing tumors and may be radiologically silent. Due to high vascularity of these lesions, it may be very difficult to manage associated bleeding. A sound knowledge of vascular anatomy of this area is important in managing intraoperative complications. Until now, only a few cases of ethmoidal hemangiomas have been reported in association with concha bullosa. Screening for these lesions may be important in patients with concha bullosa.

Conclusion: Sound knowledge of surgical anatomy forms the basis of managing intraoperative complications in endoscopic sinus surgery. Further research should be carried out to find out the association between concha bullosa and ethmoidal hemangioma and patients with concha bullosa should be screened with advanced imaging techniques for such vascular lesions where feasible.

1. Introduction

Hemangiomas are common vascular tumors of head and neck region. However, hemangiomas of the sinonasal cavity are unusual and those arising from paranasal sinuses (PNS) such as the ethmoidal sinus are rare [1]. These lesions are highly vascular and commonly visualized on radiology. We describe a case of unexpected, radiologically silent ethmoidal hemangioma which bled profusely during conchoplasty and was managed successfully by anterior ethmoidal artery cauterization via endoscopic intranasal approach. This work has been reported in line with the SCARE 2020 guidelines [2].

2. Case presentation

A 41-year-old female presented to our teaching hospital with progressive difficulty in nose breathing and associated headache since two years. She also complained of recurrent pharyngitis. There was no history of similar illness in the family and past medical as well as surgical history was non-revealing. Examination revealed decreased airflow in the left nostril when compared to right. Through a nasal endoscopic examination, we could visualize right deviated nasal septum (DNS). Computed tomography (CT) scan of nose and PNS showed right DNS with left concha bullosa (CB) (Fig. 1). Our patient was planned for endoscopic septoplasty with left conchoplasty under general anesthesia. The procedure was carried out by a specialist head and neck surgeon with a decade of experience alongside a second year head and neck surgery resident trainee. During conchoplasty, after the concha was divided vertically and lateral lamella taken out by microdebrider, sudden gush of blood filled up the nasal cavity. Temporary merocel nasal packing was done. The bleeding was so profuse that we could not visualize our surgical site. We suspected that some unusual pathology was present. On re-examination, blood was seen oozing from some

* Corresponding author.
E-mail address: rikkymikky@gmail.com (M. Karn).

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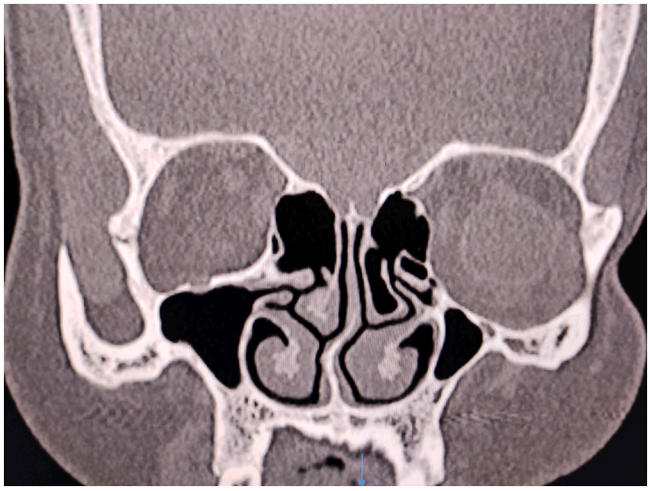

Fig. 1. Coronal CT scan showing concha bullosa on the left middle turbinate and a right DNS.

unusual tissue in the skull base lateral to medial lamella of middle turbinate and medial to lamina papyracea and there were feeder vessels coming from the anterior ethmoidal artery (AEA). Specimens from that site were sent for histopathology. When repeated measures to achieve hemostasis failed, bipolar cauterization of AEA was carried out successfully. Then, endoscopic septoplasty was performed for DNS within the next 20 min. The bleeding area was subject to repeated inspection during surgery and we found no further bleeding. There was no evidence of cerebrospinal fluid leak and orbital swelling intraoperatively. After finishing the procedure, we put in Surgicel and absorbable gelatin sponge. Finally, merocel nasal packing was done. Our patient was closely monitored in the Intensive Care Unit and the nasal pack was removed after 48 h. No complications occurred during the immediate postoperative period. Our patient was discharged postoperative day four. At two weeks follow-up, she remained symptom-free. Histopathology confirmed the bleeding lesion to be capillary hemangioma (Fig. 2).

3. Discussion

Capillary hemangiomas are composed of capillary-sized vessels lined by flat epithelial cells separated by collagen stroma. They usually arise from nasal septum, frequently affect reproductive age-group females and are commoner than their cavernous counterparts [3]. These lesions are usually small and appear as reddish-purple masses on examination. They may remain asymptomatic or present clinically with recurrent epistaxis and pain. On CT, hemangiomas appear as well-circumscribed masses with no internal calcification and homogeneous enhancement. Symptomatic sinonasal hemangiomas are managed by open or endoscopic sinus surgery with/out pre-operative embolization [4]. CB is a normal anatomic variation of middle concha characterized by its pneumatization [5]. This condition is usually asymptomatic, but symptomatic ones that cause airway obstruction or sinusitis should be managed surgically.

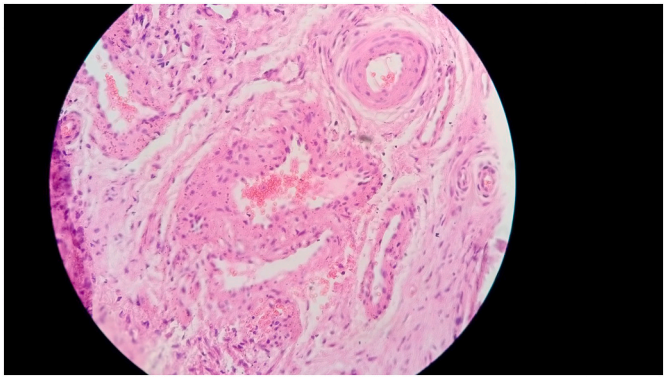

Fig. 2. Histopathology showing dilated, thin-walled vessels lined by single layer of flat endothelial cells and abundant stroma suggestive of capillary hemangioma.

In our case, the patient did not report any episode of epistaxis throughout her life. Her clinical and radiological features were consistent with CB and DNS. Pre-operative CT scans were non-suggestive of any tumorous mass. This may be due to slow growing nature of sinonasal cavity hemangiomas. Other imaging modalities such as MRI or angiography might have helped in diagnosing hemangioma pre-operatively but those were not done because there were no indications. The pathology was clear in the CT itself. Cost is a major issue in low-income countries and unnecessary tests should be avoided. In our case, we found the hemangioma was being fed from AEA. Endoscopic intranasal application of bipolar cautery to AEA successfully stopped the hemorrhage. Cauterization of AEA was done considering the hemangioma localization in ethmoidal sinus area and visualization of feeder vessels. Wherever possible, the feeder branches from the AEA should only be cauterized to prevent complications such as ischemia and resultant septal necrosis. A sound knowledge of vascular anatomy of this area is important in managing intraoperative complications. It is hypothesized that compression effect of CB may lead to vascular proliferation and hemangioma formation [6]. We recommend a thorough pre-operative evaluation for hemangioma in patients with CB. Further research should be carried out to find out any association between them.

4. Conclusion

Anatomical basis of surgical practice is essential for managing complications during surgeries around the nose and paranasal sinuses. Head and neck surgeons should be prepared for managing unexpected pathologies such as ethmoidal hemangioma during endoscopic sinus surgery. In patients with concha bullosa, attempt should be made to look for vascular lesion such as hemangiomas where advanced imaging modalities are available.

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Ethical approval

No ethical approval necessary.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Research registration

Not applicable.

Guarantor

Brihaspati Sigdel.

Disclaimer

No patient or author details are included in the figures.

Provenance and peer review

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None.

CRediT authorship contribution statement

Sigdel B and Karn M designed the report, reviewed the literature, edited the images and wrote the paper. Karn M and Kandel D analyzed the data and revised the paper. All the authors have read and approved the final manuscript.

Declaration of competing interest

The authors report no conflicts of interest.

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