Annals of Clinical Otolaryngology

Case Report
Published: 30 Jan, 2017

Case Report and Literature Review of Atypical Angiofibromas of the Inferior Turbinate

Brown LA[1]*, Joe SA[1] and Cabay RJ[2]

[1]Department of Otolaryngology/Head and Neck Surgery, University of Illinois, USA

[2]Department of Oral Medicine and Diagnostic Sciences, University of Illinois, USA

Abstract

Angiofibromas are a rare benign but aggressive head and neck tumor, most commonly originating in the nasopharynx of adolescent males. There have been many cases reported of extranasopharyngeal angiofibromas, and 10 previous cases reported of angiofribromas originating from the inferior turbinate in both females and males of a wide age range. Here we present the case of a 49 year old male with an inferior turbinate angiofibroma and review the previous literature reported on inferior turbinate angiofibromas.

Introduction

Nasopharyngeal angiofibroma (NA) area rare benign but clinically aggressive tumor most commonly found in the nasopharynx, originating from the pterygopalatine fossa [1]. Its incidence has been reported as 0.5-0.05 % of head and neck tumors [2] and is most commonly present in young males age 7-19 years old with unilateral nasal obstruction and epistaxis (thus sometimes referred to as juvenile nasopharyngeal angiofibroma) [1-3].

Various case reports have demonstrated the rare presentation of NAs in both the elderly and the very young, in females, and in locations outside of the nasopharynx [4,5]. The most common extranasopharyngeal locations are the maxillary and ethmoid sinuses [6]. There have been11 cases reported of angiofibromas originating from the inferior turbinates (ITs) in our literature review [4,7-10]. Celik et al. [4] proposes a system of grading atypical angiofibromas based on location, age, gender, presenting symptoms, histopathology and multifocality.

Here we present the unusual case of a 49 year-old male with an angiofibroma originating from the left IT, which is atypical based on age and the location of the tumor, and we review the literature of previously reported IT angiofibromas.

Case Presentation

A 49 year-old Hispanic male presented to the Otolaryngology clinic with 4-5 months of left-sided epistaxis and one month of left-sided nasal obstruction. Ared polypoid mass was visualized on nasal endoscopy, obstructing the entire nasal cavity. A biopsy was attempted in clinic butwas complicated by significant bleeding, thus he was taken to the operating room for biopsy in a controlled setting. Pathology returned asstromal fibrous tissue with vascular structures, suggestive of NA. Pre-operative computed tomography (CT) imaging showed a left sided heterogeneously enhancing expansile mass (5.9 x 2.6 x 4.0 cm) extending from the choana into the maxillary and ethmoid sinuses, and engulfing the superior, middle, and inferior turbiantes (Figure 1).

He underwent pre-operative embolization via two small branches off of the internal maxillary artery, andthree days later underwentresection. Resection was carried out using coblation and a microdebrider in addition to sharp dissection, assisted by computer image guidance. Intraoperatively, a large,vascular, friable mass was visualized filling the left nasal cavity with attachments along the entire inferior turbinate. The mass did not appear to extend into the ethmoid air cells, maxillary sinus, or pterygopalatine fossa. Pathology showed densefibrocollagenous tissue with slit-like vascular channels, consistent with NA (Figure 2).

Discussion

NAs most commonly occur in young males aged 7-19 year old,but there have been rare cases reported in malesbeyondthis age range. There have also beencases reported in females [2,4-6].

OPEN ACCESS

Correspondence:
Lisa A. Brown, Department of Otolaryngology/Head and Neck Surgery, University of Illinois at Chicago, Chicago, IL USA, Tel: 310-971-1112;
E-mail: brownlis@uic.edu

Received Date: *28 Nov 2016*
Accepted Date: *27 Jan 2017*
Published Date: *30 Jan 2017*

Citation:
Brown LA, Joe SA, Cabay RJ. Case Report and Literature Review of Atypical Angiofibromas of the Inferior Turbinate. Ann Clin Otolaryngol. 2017; 2(1): 1007.

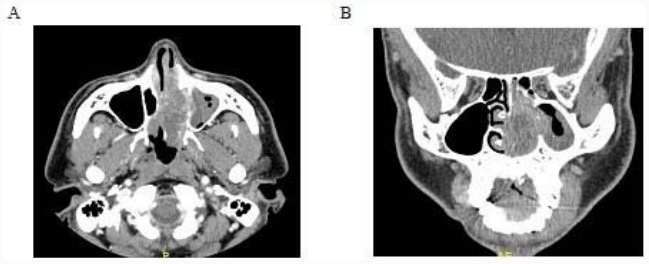

Figure 1: (A) Axial and (B) Coronal sections of CT showing left sided heterogeneously enhancing expansile mass, which extends from choanae to maxillar and ethmoid sinuses, and engulfs the superior, middle, and inferior turbinates. Intraoperatively, mass was seen to be arising from the entire inferior turbinate, and the left maxillary sinus and ethmoid sinus had no tumor extension.

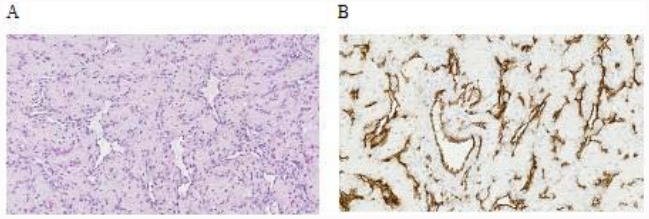

Figure 2: (A) Hematoxylin and eosin-stained section reveals fibrocollagenous stroma with slit-like vascular channels, (B) Immunohistochemical stain showing CD34 positivity in the thin layer of endothelial cells lining the vascular channels.

There is speculation that NAs are affected by hormones, such as dihydrotestosterone, which is why they are most commonly seen in young males [11]. These tumors normally arise in the pterygopalatine fossa at the aperture of the vidian canal, and grow toward the lateral wall of the nasopharynx [1,9].

Huang et al. [6] performed a literature review of 55 cases of extranasopharyngeal angiofibromas. They found that these angiofibromas are more likely to be found at a later age (22 years old) and that 25.5% were found in women. The same review reported 11% of these extranasopharyngeal angiofibromas occurred in people over the age of 50, and the most common locations were the maxillary and ethmoid sinuses, and they reported 2 cases of IT angiofibromas.

Celik et al. [4] proposed a system forgrading atypical angiofibroma based on location, age, gender, presenting symptoms, histopathology and multifocality. The above presented case meets criteria via two mechanisms: age and location.

Our literature search revealed 11 previous reports of NA originating from the IT. It is proposed that the tumor's location is secondary to ectopic tissue located in the IT. Celik et al [4] reports two cases, the first is of a 33 year-old male with nasal swelling and obstruction, and the other of a 60 year-old male with nasal polyps and an angiofibroma found on the tail of the IT. Others have reported IT angiofibromas in females aged 8, 26, 28, 52 and 78 years old, and in males aged 9, 10, 30 and 62 years old. The most common presenting symptoms were unilateral or bilateral nasal obstruction and intermittent epistaxis [6-10,12,13]. We have included our case in the below reporting of IT angiofibromas.

The mean age of presentation of the IT angiofibroma is 37.1 years, 36.1 years for males and 38.4 years for females. Our case reports a 49-year-old male with an IT angiofibroma which is above the average ageof presentation for males and females combined, and

A

Age (years)	Gender (F = female; M = male)
8	F
9	M
10	M
26	F
28	F
30	M
33	M
49	M
52	F
60	M
62	M
78	F

B

	Total (n=12) (years)	Males (n=7) (years)	Females (n=5) (years)
Mean	37.1	36.1	38.4
Median	31.5	33	28

Figure 3: (A) Demonstrates the age and gender of patients with reported IT angiofibromas, (B) Demonstrates the mean and median of the patients with IT angiofibromas, separated also by gender.

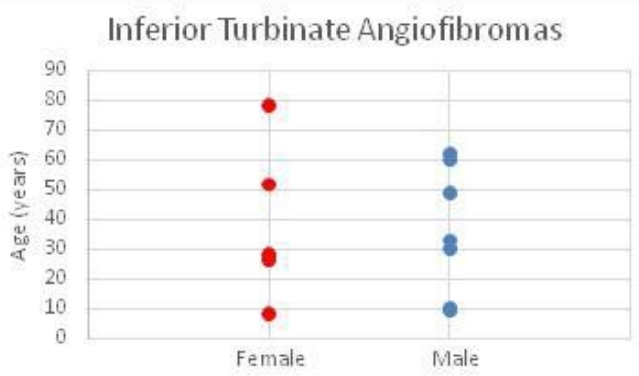

Figure 4: Graph points demonstrating distribution across age and gender of patients with reported IT angiofibromas.

males alone (Figure 3). Our case also exhibits a more extensive tumor involvement of the IT than these reported cases. The incidence of reported IT angiofibromas is slightly higher in males than females (59% males, 41% females). This goes along with the previous findings that extranasopharyngeal angiofibromas have a higher incidence in females and a higher age of presentation than typical angiofibromas of the nasopharynx. However, 41% is significantly higher than the previously reported 25% of all extranasopharyngeal angiofibromas occurring in females. The average age of presentation is also significantly higher in IT versus the previously reported extranasopharyngeal angiofibromas (37.1 years vs. 22 years).

The most common causes of nasal masses are allergic or inflammatory polyps followed by hemangiomas and inverted papillomas. Greater than 90% of malignant nasal masses are squamous cell carcinoma [14]. In an elderly male, it is easy to confuse a NA for a malignant lesion due to the rarity of an NA, especially if it is outside

of the nasopharynx. However, a blue or red hue is highly suggestive of a vascular origin. Hemangiomas are more common than NAs in the nasopharynx, but both should be kept in the differential until pathologic confirmation of a diagnosis [2,6,15]. It is recommended to be cautious in regards to the biopsy of a mass that appears vascular, especially in older adults,due to the higher risk of complications secondary to bleeding and the common use of antiplatelets or anticoagulants in this population [16]. Although biopsy is usually not needed, it is suggested to perform a biopsy, if deemed necessary, in a controlled setting, such as an operating room (as we did).

Conclusion

Although angiofibromas are most commonly found in the nasopharynx of adolescent males, they also occur in extranasopharyngeal areas such as the IT. IT angiofibromas are rare with 10 cases reported previously in literature. They are more likely to be found at an older age (average 37.1 years) and with a higher incidence found in females (41.6%) than NAs and other extranasopharyneal angiofibromas. An IT angiofibroma should be kept in the differential diagnosis of a vascular-appearing nasal mass originating from the IT in both males and females.

Acknowledgement

Andre Kajdacsy-Balla, MD, PhD, and Nasma Majeed, MBChB from the University of Illinois Hospital & Health Science System Pathology department.

References

1. Lloyd G, Howard D, Phelps P, Cheesman A. Juvenile angiofibroma: the lessons of 20 years of modern imaging. J Laryngol Otol. 1999; 113: 127-134.
2. Gullane PG, Davidson J, O'Dwyer T, Forte V. Juvenile angiofibroma: a review of the literature and a case series report. Laryngoscope. 1992; 102: 928-933.
3. Johnson J. Bailey's Head and Neck Surgery: Otolaryngology. 5th ed. Philadelphia: Wolters Kluwer; 2015.
4. Celik B, Erisen L, Saraydaroglu O, Coskun H. Atypical angiofibromas: a report of four cases. Int J Pediatr Otorhinolaryngol. 2005; 69: 415-421.
5. Patrocinio JA, Patrocinio LG, Borba BH, Bonatti BDe S, Guimarães AB. Nasopharyngeal angiofibroma in an elderly woman. Am J Otolaryngol. 2005; 26: 198-200.
6. Huang RY, Damrose EJ, Blackwell KE, Cohen AN, Calcaterra TC. Extranasopharyngeal angiofibroma. Int J Pediatr Otorhinolaryngol. 2000; 56: 59-64.
7. Nomura K, Shimomura A, Awataguchi T, Murakami K, Kobayashi T. A case of angiofibroma originating from the inferior nasal turbinate. Auris Nasus Larynx. 2006; 33: 191-193.
8. Baptista MAFB, Pinna FdR, Voegels RL. Extranasopharyngeal Angiofibroma Originating in the Inferior Turbinate: A Distinct Clinical Entity at an Unusual Site. Int Arch Otorhinolaryngol. 2014; 18: 403-405.
9. Maheshwarappa RP, Gupta A, Bansal J, Kattimani MV, Shabadi SS, Baser SC. An unusual location of juvenile angiofibroma: a case report and review of the literature. Case Rep Otolaryngol. 2013; 2013: 175326.
10. Salimov A, Ozer S. A Rare Location of Angiofibroma in the Inferior Turbinate in Young Woman. Int Arch Otorhinolaryngol. 2015; 19: 187-190.
11. Farag MM, Ghanimah SE, Ragaie A, Saleem TH. Hormonal receptors in juvenile nasopharyngeal angiofibroma. Laryngoscope. 1987; 97: 208-211.
12. Singh GB, Agarwal S, Arora R, Doloi P, Kumar D. A Rare Case of Angiofibroma Arising from Inferior Turbinate in a Female. J Clin Diagn Res. 2016; 10: MD07- MD08.
13. Taggarshe D, Quraishi MS, Dugar JM. Inferior turbinate angiofibroma: an atypical presentation [correction of preservation]. Rhinology. 2004; 42: 45-47.
14. Lathi A, Syed MMA, Kalakoti P, Qutub D, Kishve SP. Clinico-pathological profile of sinonasal masses: a study from a tertiary care hospital of India. Acta Otorhinolaryngol Ital. 2011; 31: 372-377.
15. Tiwari PK, Teron P, Saikia N, Saikia HP, Bhuyan UT, Das D. Juvenile Nasopharyngeal Angiofibroma: A Rise in Incidence. Indian J Otolaryngol Head Neck Surg. 2016; 68: 141-148.
16. U.S. Spends Nearly $1 Billion on Blood Thinners for Adults. US Department of Health and Human Services 353. 2010; 1-2.