CASE REPORT

Journal of Otorhinolaryngology and Facial Plastic Surgery. 2023;9(1):1-4.

https://doi.org/10.22037/orlfps.v9i1.44881

Nasopharyngeal Inflammatory Pseudotumor: A Case Presenting with Nasal Congestion and Hearing Disorder

Farzin Davoodi[1,2], Amirreza Zaker[1,2], Narges Bazgir[1], Reza Naseri[3*], Farahnaz Bidari Zerehpoosh[4], Sana Sheikhaghayi[5], Mohsen Fazli[1,2]

1. Hearing Disorders Research Center, Loghman Hakim Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
2. Department ofOtorhinolaryngology, Loghman Hakim Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
3. Department of Radiology, Loghman Hakim Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
4. Department of Pathology, Loghman Hakim Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
5. Student Research Committee, Shahid Beheshti University of Medical Sciences, Tehran, Iran.

Article Info

Article Note:
Received: November 2023
Accepted: December 2023
Publish Online: December 2023

***Corresponding Author**:
Dr Reza Naseri

Email:
dr.rezanaseri@sbmu.ac.ir

Keywords:
Sinonasal;
Inflammatory pseudotumor;
Hearing disorder;
Nasal congestion.

Abstract

Background: Inflammatory pseudotumor (IP) is a collection of histocytes, and inflammatory cells including plasma cells, lymphocytes, eosinophils, and fibroblasts. The IP in the nasopharyngeal space is exceedingly rare. If left untreated it may cause permanent and serious complications.

Case presentation: A 46-year-old man with left nasal congestion, and a hearing decrement in his left ear was admitted to Loghman-hakim Hospital. In audiometric evaluations, there was a 20 decibels gap in the hearing threshold between two ears. In nasopharyngoscopy, there was a large gray-to-white mass with intact epithelia in the nasopharynx. In a computed tomography scan, a large soft mass in the left nasopharynx, with no evidence of bone destruction. Magnetic resonance imaging (MRI) homogenous isointense and hypointense mass in T1-weighted and T2-weighted images, respectively. Based on all the investigations, the risk of malignancy was considered low. As a result, the final diagnosis of IP was considered for him.

Discussion: IP can occur anywhere in the body and encompasses a diverse range of masses, including inflammatory myofibroblastic tumors, IPTs of lymph nodes, orbit, and spleen, genitourinary pseudosarcomatous, and post-infectious or reparative disorders. The tumors usually affect children and young adults, and the precise incidence and prevalence of IPs are difficult to estimate. The main treatment is surgical resection, corticosteroids, or both, with alternative treatments including small molecule inhibitors, immunoglobulin, and radiotherapy.

Conclusion: Sinonasal IPs are exceedingly rare. In this article, we reported a mid-aged man with an IP in his left nasopharynx space.

Conflict of Interest: The authors declare no conflict of interest.
Please cite this article as: Davoodi F, Zaker A, Bazgir N, Naseri R, Bidari Zerehpoosh F, Sheikhaghayi S, Fazli M. Nasopharyngeal Inflammatory Pseudotumor: A Case Presenting with Nasal Congestion and Hearing Disorder. J Otorhinolaryngol Facial Plast Surg. 2023;9(1):1-4. https://doi.org/10.22037/orlfps.v9i1.44881

Introduction

Inflammatory pseudotumor (IP) is a collection of histocytes, and inflammatory cells including plasma cells, lymphocytes, eosinophils, and fibroblasts (1). Various terms such as plasma cell granuloma, histiocytoma complex, fibrous xanthoma, inflammatory myofibroblastic tumors and other terms are used for IP (2). The most common place for IPs to arise is the lung. The prevalence of head and neck IP accounts for about five percent (2). The IP in nasopharyngeal space is exceedingly rare. If left untreated it may cause permanent and serious complications. The most frequent clinical manifestations of IPs in the nasopharyngeal cavity and sinuses consist of nasal congestion, and epistaxis (3). In this

Journal of Otorhinolaryngology and Facial Plastic Surgery. 2023;9(1):1-4.
https://doi.org/10.22037/orlfps.v9i1.44881

article, we describe a case with an IP in his left nasopharyngeal space.

Case Presentation

A 46-year-old man with snoring left nasal congestion and a hearing decrement in his left ear was admitted to Loghman-hakim Hospital. He developed his symptoms over the past three months. During this period, his nasal congestion remained constant. His past medical and surgical history was insignificant. Except for left nasal congestion and the presence of fluid in his left middle ear, physical examinations were otherwise normal. In audiometric evaluations, there was a 20 decibels gap in the hearing threshold between two ears. In nasopharyngoscopy, there was a large gray-to-white mass with intact epithelia in the nasopharynx. In computed tomography (CT) scan a large soft mass in the left nasopharynx, with no evidence of bone destruction (Figure 1). In imaging evaluations, the mass was extended from the skull base to the nasopharynx.

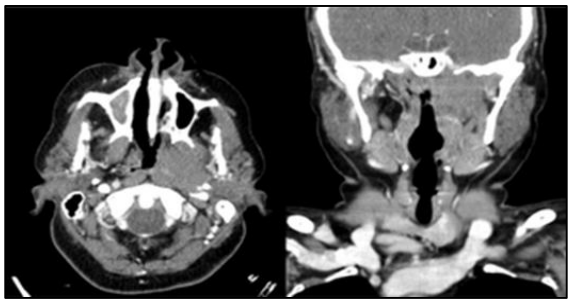

Figure 1. The axial and coronal CT of the patient.

Magnetic resonance imaging (MRI) homogenous isointense and hypointense mass in T1-weighted and T2-weighted images, respectively (Figure 2, 3).

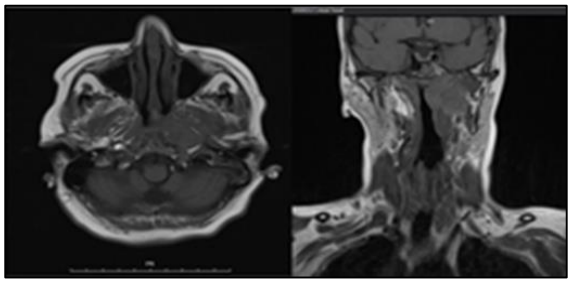

Figure 2. The axial and coronal T1-weighted MRI of the patient.

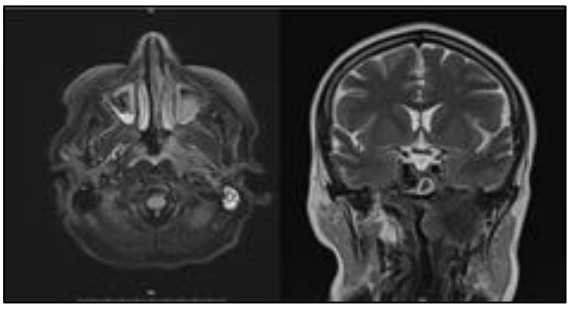

Figure 3. The axial and coronal T2-weighted MRI of the patient.

The mass was biopsied and the specimen was sent to pathology. In pathological evaluations, nasopharyngeal mucosa with reactive follicular hyperplasia and infiltration of skeletal and fibrotic tissue with mixed inflammatory cells were evident. All the mentioned changes were suggestive of inflammatory pseudotumor (IP). In histological examination, the superficial specimen showed respiratory mucosa with chronic infiltration and fibrosis. In a deep biopsy, chronic inflammatory cells infiltrate in densely fibrotic tissue (Figure 4).

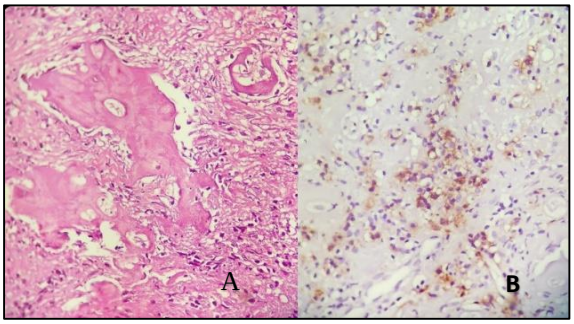

Figure 4. A: Destruction of hyalin cartilagewithin tumoral tissue, H&E stain,x400 B: (Immunohistochemistry staining) CD138 stained plasma cells,x400

On immunohistochemistry (IHC) staining CD138, kappa, and lambda were positive, Des and S 100 were negative. Based on IHC staining the probability of IgG 4-related IP was considered low. Based on all the investigations, the risk of malignancy was considered low. As a result, the final diagnosis of IP was considered for him. After receiving treatment, the mass was resolved completely and no evidence of recurrence was observed (Figure 5).

Journal of Otorhinolaryngology and Facial Plastic Surgery. 2023;9(1):1-4.
https://doi.org/10.22037/orlfps.v9i1.44881

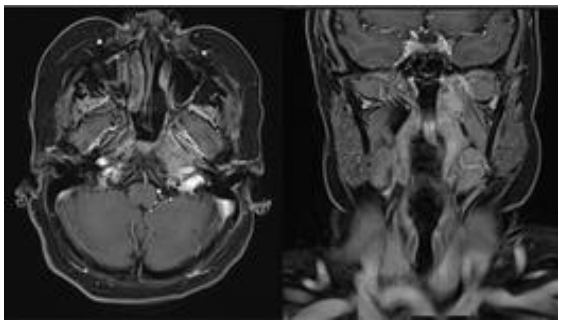

Figure 5. The axial and coronal MRI of the patient after receiving treatment.

Discussion

Inflammatory pseudotumor (IP) was first described in 1903 by Gleason and Busse, and then it was categorized as a different entity in 1905(4). While it is mostly described in the literature as soft tissues in the lungs, it occurs anywhere in the body, from the head and neck to the pelvis (2). IP consists of a wide variety of masses including inflammatory myofibroblastic tumors (IPT), IPTs of lymph nodes, orbit, and spleen, genitourinary pseudosarcomatous, and post-infectious or reparative disorders. In histopathological investigations, infiltrations of inflammatory cells such as lymphocytes, histocytes, plasma cells, and abundant numbers of fibroblasts and myofibroblasts (2).

These tumors typically affect children and young adults. But The prevalence of sinonasal IPs does not favor any particular age(2). Due to unclear definitions, the precise incidence and prevalence of IPs are hard to estimate. The majority of IPs are located in the lungs and only fifteen percent of them occur in the head and neck region (2). Extra-orbital IPs usually occur in the nasopharynx, maxillary sinus, larynx, and nasal cavity trachea (2).

IPs are the diagnosis of exclusion and are related to idiopathic inflammation. Trauma, foreign body, and infections especially with mycobacteria, *Rhodococcus equi*, and *Klebsiella rhinoscleromatis* (5).

The clinical course of IPs is usually benign, recurrences and aggressive behaviors have been described, though (6). Clinical presentations of sinonasal IPs include bone destruction, and a good prognosis (3). The presented case had nasal congestion, snoring, and a decrease in hearing threshold. No evidence of bone destruction was observed.

In sinonasal IPs, bony changes are evident on CT scans (3). We observed no bone erosion, sclerosis, or remodeling on the CT scan of the presented case. On MRI evaluations, sinonasal IPs are usually iso/hypointense in T1-weighted and hypointense in T2-weighted MRI (7). Our case had the same pattern in MRI.

On pathological evaluations, the presence of spindle cells, plasma cells, and lymphocytes is evident. Other cells such as myofibroblasts, fibroblasts, histocytes, and inflammatory filtrates may be present (5). On IHC, smooth muscle actin, cytokeratin, CD20, and anaplastic lymphoma kinase are usually positive (5). In the presented case, CD138, kappa, and lambda were positive on IHC staining.

The main treatment of IPs is surgical resection, corticosteroids, or both. Another alternative treatment composes of small molecule inhibitors, immunoglobulin, and radiotherapy (8).

Conclusion

In this article, we reported a mid-aged man with nasal congestion, hearing decrement, and snoring. After various investigations, he was finally diagnosed with IP in his left nasopharynx.

Acknowledgements

We would like to thank anyone who contributed to this study.

Patient Consent

Written consent was obtained from the patient. The whole process of examination and the purpose of the article was thoroughly explained.

Conflict of Interest

The authors declared no conflicts of interest.

Journal of Otorhinolaryngology and Facial Plastic Surgery. 2023;9(1):1-4.

https://doi.org/10.22037/orlfps.v9i1.44881

Financial Support

The authors declared that there was no financial support.

Author contribution

RN conceptualized the study, FD acquisition of data, NB and FD drafting the manuscript, NB, RN revising for critical intellectual concept and approved of the version to be submit.

Authors' ORCHIDs

Farzin Davoodi
https://orcid.org/0000-0003-4609-3460
Narges Bazgir
https://orcid.org/0000-0002-6443-9448
Reza Naseri
https://orcid.org/0000-0003-4593-124X
Farahnaz Bidari Zerehpoosh
https://orcid.org/0000-0002-7817-4938

References

1. Huang WH, Dai YC. Inflammatory pseudotumor of the nasal cavity. Am J Otolaryngol. 2006 Jul-Aug;27(4):275-7. PubMed PMID: 16798408. Epub 2006/06/27. eng. Web of Science

2. Coffin CM, Watterson J, Priest JR, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. Am J Surg Pathol. 1995 Aug;19(8):859-72. PubMed PMID: 7611533. Epub 1995/08/01. eng. Web of Science

3. Li J, Wang M, Li W, Tao Y, Shi X. Inflammatory Pseudotumor in the Nasal Cavity and Sinuses: A Case Report and Associated Literature Review. Ear Nose Throat J. 2021 Dec;100(10_suppl):897S-901S. PubMed PMID: 32419496. Epub 2020/05/19. eng.

4. Ortlip TE, Drake VE, Raghavan P, Papadimitriou JC, Porter NC, Eisenman DJ, et al. Inflammatory Pseudotumor of the Temporal Bone: A Case Series. Otol Neurotol. 2017 Aug;38(7):1024-31. PubMed PMID: 28570415. Pubmed Central PMCID: PMC5762198. Epub 2017/06/02. eng.

5. Devaney KO, Lafeir DJ, Triantafyllou A, Mendenhall WM, Woolgar JA, Rinaldo A, et al. Inflammatory myofibroblastic tumors of the head and neck: evaluation of clinicopathologic and prognostic features. Eur Arch Otorhinolaryngol. 2012 Dec;269(12):2461-5. PubMed PMID: 22588194. Epub 2012/05/17. eng.

6. Gale N, Zidar N, Podboj J, Volavsek M, Luzar B. Inflammatory myofibroblastic tumour of paranasal sinuses with fatal outcome: reactive lesion or tumour? J Clin Pathol. 2003 Sep;56(9):715-7. PubMed PMID: 12944561. Pubmed Central PMCID: PMC1770050. Epub 2003/08/29. eng.

7. Maruya S, Miura K, Tada Y, Masubuchi T, Nakamura N, Fushimi C, et al. Inflammatory pseudotumor of the parapharyngeal space: a case report. Auris Nasus Larynx. 2010 Jun;37(3):397-400. PubMed PMID: 19857937. Epub 2009/10/28. eng.

8. Kansara S, Bell D, Johnson J, Zafereo M. Head and neck inflammatory pseudotumor: Case series and review of the literature. Neuroradiol J. 2016 Dec;29(6):440-6. PubMed PMID: 27650653. Pubmed Central PMCID: PMC5131759. Epub 2016/09/22. eng.