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A Case of Nasal Myiasis in a Patient with Intestinal Type Sinonasal Adenocarcinoma

İntestinal Tip Sinonazal Adenokarsinomlu Bir Hastada Nazal Miyazis Olgusu

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ABSTRACT Myiasis is an infestation caused by fly, mosquito or midge larvae settling on animals or humans and feeding on dead or living tissues or nutrients digested by them. Myiasis of the nasal cavity is an uncommon entity. It may particularly be encountered in individuals with low socioeconomic levels in need of care or against a background of malignancy, but is very rare in healthy individuals. While there is no clear algorithm, daily endoscopy-guided debridement with saline irrigation will be beneficial in treatment. We report a case of nasal myiasis in an 82-year-old male patient reported as intestinal type sinonasal adenocarcinoma following biopsy taken from a mass completely filling the nasal cavity two years previously, but who had received no treatment during that time.

Keywords: Adenocarcinoma; myiasis; nasal cavity; nasal obstruction

Myiasis is an infestation caused by fly, mosquito or midge larvae settling on animals or humans and feeding on dead or living tissues or nutrients digested by them. The manifestation caused by larvae settling in living tissues is known as primary myiasis, while that caused by larvae in dead tissues is known as secondary myiasis. Nasal, ocular, oral, gastrointestinal urogenital region, and cutaneous involvement may be observed. While it is very rare in healthy individuals, myiasis may be seen in individuals with low socioÖZET Miyazis; sinek, sivrisinek, tatarcık larvalarının hayvan veya insanlara yerleşerek, onların ölü veya canlı dokularıyla veya sindirdikleri besinlerle beslenerek yaptıkları infestasyondur. Nazal kavitenin miyazisi, sık görülen bir durum değildir. Özellikle sosyoekonomik düzeyi yetersiz, bakıma muhtaç kişilerde ve malignite zemininde bu durumla karşılaşılabilmektedir. Sağlıklı kişilerde görülmesi oldukça nadirdir. Tedavisinde net bir algoritma olmamakla beraber salinli irrigasyon ve endoskopi eşliğinde günlük debridman yapılması fayda sağlayacaktır. Burada, 2 sene önce nazal kaviteyi tamamen dolduran kitleden alınan biyopsi sonucu intestinal tip sinonazal adenokarsinom olarak raporlanan, ancak bu süreçte herhangi bir tedavi almamış olan 82 yaşındaki erkek hastada gelişen nazal miyazis olgusunu sunduk.

Anahtar Kelimeler: Adenokarsinom; miyazis; nazal kavite; nazal tıkanıklık

cultural levels, due to poor hygienic conditions, or in individuals with an underlying malignancy or disease.¹ We report a case of nasal myiasis developing in an 82-year-old man with untreated sinonasal adenocarcinoma.

CASE REPORT

Written informed consent was obtained from the patient who participated in this case. An 82-year-old

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1307-7384 / Copyright © 2022 Turkey Association of Society of Ear Nose Throat and Head Neck Surgery. Production and hosting by Türkiye Klinikleri. This is an open access article under the CC BY-NC-ND license (https://creativecommons.org/licenses/by-nc-nd/4.0/). man presented to our clinic due to widespread larvae filling the bilateral nasal cavities. The patient's history revealed that he had presented to a physician two years previously due to nasal obstruction and headache, and that intestinal type sinonasal adenocarcinoma had been reported following biopsy from a mass completely filling the left nasal cavity. The patient failed to attend subsequent check-ups and had received no treatment. Endoscopic examination revealed a vegetant mass and larvae completely filling the bilateral nasal cavities, and lateral displacement of the left orbita caused by the mass (Figure 1A, Figure 1B). Cerebral and orbital computed tomography revealed a mass with indistinct margins, approximately 67x42 mm in size, causing destruction in the surrounding osseous structures and completely filling the bilateral nasal cavities (Figure 2). The patient was admitted to our clinic and was started on parenteral antibiotherapy. Saline irrigation and rigid endoscopeguided debridement were performed. No larvae were observed in the nasal cavity at the end of the third day. Positron emission tomography/computed tomography was performed, and the patient was transferred to the radiation oncology clinic. The size of the mass decreased after radiotherapy. But the patient who was being followed up by the radiation oncology clinic died 6 months later.

DISCUSSION

Sinonasal malignancies are rare entities constituting approximately 3% of upper respiratory tract malignancies. Initial symptoms are identical to benign lesions and inflammatory diseases.² Squamous cell carcinoma is the most common malignant tumor of the sinonasal tract.³ However, sinonasal adenocarcinomas are rare malignancies, and constitute only 0.4% of neoplasms in humans. Sinonasal adenocarcinomas are more frequently seen in individuals exposed to timber, leather and textile dusts, nickel, dye, and varnish.⁴ Tobacco use is also associated with sinonasal squamous cell carcinoma, but its relationship with adenocarcinoma is weak.⁵ Sinonasal adenocarcinomas originate from the respiratory surface epithelium and the seromucinous gland beneath. They may exhibit a growth pattern resembling that of carcinomas or adenomas of intestinal origin, or may mimic the normal histology of the intestinal mucosa. They are classified as "intestinal" or "non-intestinal" depending on their resemblance to the intestinal mucosa. Intestinal type adenocarcinoma is the second most common form of sinonasal adenocarcinoma, after adenoid cystic carcinoma. Intestinal adenocarcinomas are more common in men, and the mean age at appearance is 50-64 years. Common initial symptoms include nasal obstruction, epistaxis and rhinor-



FIGURE 1: A) Larvae completely filing the nasal cavity. B) Larvae removed from the nasal cavity.



FIGURE 2: Computed tomography image of the mass in the nasal cavity.

rhea. They are most frequently located in the ethmoid sinus, the nasal cavity, and the maxillary antrum. These are aggressive tumors that can also cause local spread to the orbita, the base of the skull and the intracranial cavities, and distant metastasis.⁶ Metastatic adenocarcinomas of the gastrointestinal tract must also be excluded using appropriate imaging techniques or endoscopy at diagnosis.⁷

Myiasis of the nasal cavity is an uncommon entity. Several species of fly are capable of causing myiasis in humans. While myiasis is rare worldwide, it is more common in developing countries, in warm humid climates, and in communities with poor or inadequate health and hygiene conditions. The risk of myiasis is also known to be higher in unhealthy, immunosuppressed individuals and against a background of malignancy. Presentation symptoms among patients with myiasis may vary. In the early period, patients may present with sneezing and nasal discharge. However, the most common presentation symptoms are headache, facial pain, purulent nasal discharge, and larvae in the nose. Extensive tissue damage may occur in the nasal cavity and sinuses, secondary bacterial infections may develop, and meningitis-related mortality may be seen.⁸ Severe allergic reaction associated with nasal myiasis has also been reported.¹ Identification of species causing myiasis is a task for entomologists. The larvae are sent for analysis after being removed from the nasal cavity and fixed in ethanol.9 No such analysis was possible in our case since the requisite technical means

were unavailable. There is no clear algorithm in the treatment of myiasis, and local and systemic methods are applied together. Saline irrigation and endoscopy-guided manual debridement will be beneficial. Since larvae are photophobic, they tend to settle in dark regions, and this makes extraction more difficult. Nasal endoscopy permits easy visualization of the larvae, and thus assists treatment. Turpentine oil and liquid paraffin drops have been reported to be effective in myiasis treatment.8 Turpentine oil destroys the larvae, while liquid paraffin cuts off their source of oxygen, thus forcing them outside. Systemic antibiotherapy should also be initiated in order to prevent secondary bacterial infections that may develop in association with tissue damage. Raising environmental standards also plays an important role in controlling the fly population and myiasis.¹⁰

In conclusion, myiasis appears that patients having severe ill, poor hygienic condition and predisposed diseases. Nasal cavity is a possible site for myiasis. Physicians must be aware of myiasis especially in these patients.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Fatma Atalay; Design: Ayhan Kars; Control/Supervision: Ayhan Kars; Data Collection and/or Processing: Sinan Köyceğiz; Analysis and/or Interpretation: Fatma Atalay; Literature Review: Atahan Ağrılı; Writing the Article: Fatma Atalay; Critical Review: Mustafa Sıtkı Gözeler; References and Fundings: Mustafa Sıtkı Gözeler; Materials: Sinan Köyceğiz.

REFERENCES

- Erenler AK, Turan AP, Ay ÖO, Taylan Özkan A. Report of a rare case of severe allergic reaction due to nasal myiasis and a brief review of the literature in Turkey. SAGE Open Med Case Rep. 2019;7:2050313X19843390. [Crossref] [Pubmed] [PMC]
- Thorup C, Sebbesen L, Danø H, Leetmaa M, Andersen M, Buchwald C, et al. Carcinoma of the nasal cavity and paranasal sinuses in Denmark 1995-2004. Acta Oncol. 2010;49(3):389-94. [Crossref] [Pubmed]
- Gras Cabrerizo JR, Sarandeses García A, Montserrat I Gili JR, Orús Dotú C. [Revision of carcinomas in paranasal sinuses]. Acta Otorrinolaringol Esp. 2007;58(6):266-75. [Crossref] [Pubmed]
- Kurt B, Celik E, Inan K, Durmaz A, Kutlay M, Gunhan O. A case of a sinonasal adenocarcinoma with metaplastic ossification. B-ENT. 2016;12(3):241-4. [Pubmed]
- Nissen MS, Stokholm ZA, Christensen MS, Schlünssen V, Vestergaard JM, Iversen IB, et al. Sinonasal adenocarcinoma following styrene exposure in the reinforced plastics industry. Occup Environ Med. 2018;75(6):412-4. [Crossref] [Pubmed] [PMC]
- Leivo I. Sinonasal adenocarcinoma: update on classification, immunophenotype and molecular features. Head Neck Pathol. 2016;10(1):68-74. [Crossref] [Pubmed] [PMC]
- Kim J, Chang H, Jeong EC. Sinonasal intestinal-type adenocarcinoma in the frontal sinus. Arch Craniofac Surg. 2018;19(3):210-3. [Crossref] [Pubmed] [PMC]
- Manjunath NM, Pinto PM. Management of recurrent rhinomaxillary mucormycosis and nasal myiasis in an uncontrolled diabetic patient: a systematic approach. Int J Appl Basic Med Res. 2018;8(2):122-5. [Crossref] [Pubmed] [PMC]
- Francesconi F, Lupi O. Myiasis. Clin Microbiol Rev. 2012;25(1):79-105. [Crossref] [Pubmed] [PMC]
- Sabarigirish K, Nithya V, Saxena S, Dutta A. Nasal myiasis by Chrysomya bezziana. Med J Armed Forces India. 2018;74(1):82-4. [Crossref] [Pubmed] [PMC]