Rhinomyiasis: clinical and surgical management

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SUMMARY

Myiasis by *Oestrus ovis*, the zoonotic infestation with Diptera larvae, primarily diagnosed in goats and rams in tropical and Mediterranean countries, is an uncommon disease in humans; indeed, literature data are still lacking. Nevertheless, few cases of human myiasis have been reported, leading to benign or severe complications. Here, we report a rare case of human rhinomyiasis detected in Northern Italy. A 39-year-old Italian woman, returning from vacation in Corsica, showed several sinusal symptoms and progressive asthenia and was therefore admitted at the Otorhinolaryngology Unit of Biella Hospital, Italy. Endoscopic examination of the nasal cavity revealed some formations, morphologically identified as *O. ovis* larvae. The patient then underwent endoscopic sinus surgery, followed by complete resolution of symptoms. Clinical presentation, diagnostic work-up and therapeutic procedures have been compared with few other cases found in the literature.

BACKGROUND

Rhinomyiasis, even if rare, should be considered in travellers returning from holidays in tropical or rural Mediterranean regions, presenting with rhinosinusitis-like symptoms, not regressing with usual therapies. Prompt diagnosis and exclusion of ocular and rhinosinusal complications allow for simple and decisive surgical therapy.

CASE PRESENTATION

Myiasis is a parasitic infection caused by *Oestrus ovis*, primarily diagnosed in goats and rams in tropical and Mediterranean countries. Humans, different from sheep, are rarely affected by myiasis. The most frequent manifestation of the disease occurs as a furuncular lesion on the skin of visitors from South America and Africa.[1 2] Mucosal surfaces of the nose, oral cavity and oropharynx are less likely to be affected by parasitic infestation; however, to date, few cases have been reported in the literature.[1 3–9] On the contrary, eyes are more likely to be affected.[10 11]

All the reported cases have been observed in patients who had travelled or lived in rural areas where cattle or South American llamas are raised. As for ophthalmomyiasis, the nasal infection can result from parasitic fly's deposition in or around the nostrils, although the hypothesis of contamination through infected fingers has also been proposed.

Patients usually have symptoms, including a sensation of a small foreign body being struck in the nose and sore throat, sometimes leading to cough, nasal discharge, sneezing, laryngospasm, dyspnoea and stridor.[12]

In some cases, myiasis may lead to major complications (orbital, intracranic, palatal, etc), owing to the larvae releasing toxins that destroy the host tissue.[13–15]

Here, we describe a rare case detected in Northern Italy (Ospedale degli Infermi, Ponderano, Biella, Italy) and compare the clinical presentation, diagnostic work-up, and therapeutic and surgical procedures with few other cases found in the literature. The morphological and genetic characteristics have been analysed by the Parasitology Laboratory and by the Zooprophylactic Institute.[16]

A 39-year-old Italian woman with a clinical history of rhinoseptoplasty and a major depressive episode spent 2 weeks of vacation in Corsica in summer 2018. During her stay, she performed outdoor activities, with subsequent appearance of unilateral nasal respiratory obstruction, cough and pharyngodynia.

On her return, she was visited by her physician who prescribed her antibiotic therapy. Owing to the persistence of symptoms, she was then admitted to the Otorhinolaryngology Unit of Biella Hospital, showing mucous rhinorrhoea, frontal cephalgia with less maxillary involvement, olfactory disorders and progressive asthenia; fever was never reported.

Fibre-optic endoscopy of the patient's nose revealed some dark-edged formations, 1 mm in size, at the level of the left middle nasal meatus and near the eustachian tube. The formations, at a closer inspection, seemed to move slowly; therefore, a parasitic infection was suspected, and sampling, under endoscopic guidance, was performed.

The analysis, performed by the Parasitology Laboratory of IZS (Istituto Zooprofilattico Sperimentale), confirmed the morphological identification of *O. ovis* (figure 1). Then, a specimen was sent to the Laboratory of Genetics and Immunobiochemistry of the Istituto Zooprofilattico Sperimentale del Piemonte, Liguria e Valle d'Aosta for genetic identification using the forensically informative nucleotide sequencing method.[16] Genomic DNA was extracted from the specimen and a portion of the *Cox*1 gene was amplified by PCR reaction. Genetic analysis confirmed previous morphological identification.

Instrumental investigations based on clinical results were performed in the emergency department. Blood chemistry tests showed mild neutrophilia and C reactive protein levels <3 µg/mL. The chest X-ray showed no pulmonary thickening but a modest bilateral reinforcement of the interstitial texture, compatible with mild bronchitis.

Non-contrast maxillofacial CT scan showed bilaterally phlogistic material at the level of the anterior and posterior ethmoid and at the level of the ostiomeatal complex. On the contrary, no evidence of

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Figure 1 Microscopic image of *Oestrus ovis*.

frontal, maxillary and sphenoid sinus involvement was detected (figure 2).

Endoscopic examination and CT scan allowed the exclusion of sinonasal malignancies, atrophic rhinitis and neglected nasal foreign bodies.

The patient additionally underwent an eye examination to exclude the simultaneous presence of larvae at the ocular level. No deposits of larvae were identified on the ocular surface, stimulated with high-intensity light and analysed up to the level of fornices; the conjunctiva appeared pink, the cornea transparent and mirroring, the aqueous humour optically empty, and the optic disc pink with neat edges, regular vases and macula. The retina was stimulated in each section through the ophthalmoscope light, with no evidence of subretinal movements due to the presence of larvae.

Figure 2 Non-contrast maxillofacial CT scan showing bilaterally phlogistic material at the level of the anterior and posterior ethmoid and at the level of the ostiomeatal complex.

TREATMENT

The patient was submitted to antibiotic therapy, prescribed on the grounds of infectious disease diagnosis to treat the bronchitis (ciprofloxacin 500 mg every 12 hours+amoxicillin/clavulanate 1 g every 8 hours for 10 days); no specific antiparasitic therapy was administered considering the side effects (nausea, vomiting, diarrhoea, abdominal pain, headache).

Once CT scan and infectious and ophthalmological evaluations were performed, the patient urgently underwent endoscopic sinus surgery under general anaesthesia.

During the surgery, several larvae were identified inside the nasal cavities (nasal septum, lower and middle nasal meatus) and then removed, whereas no larvae were found at the level of the nasopharyngeal cavity and tubes. The maxillary sinus mucosa was moderately hypertrophic, hyperaemic and easily bleeding. A lower uncinectomy and bilateral meatotomy were performed; the maxillary cavities, explored with angled fibre-optic endoscope, appeared empty. Finally, the maxillary and ethmoidal sinuses were washed with 0.9% saline solution.

OUTCOME AND FOLLOW-UP

No postoperative complications were observed.

Antibiotic therapy was extended for 10 days to prevent rhinosinusal overinfections. Symptoms rapidly regressed, and no recurrence of parasitic infection has been detected during the periodic (after 2 weeks, 1, 3 and 6 months, and 1 year) clinical and instrumental follow-ups with fibre-optic endoscope.

DISCUSSION

The larvae of various Diptera species are the main causative agent of human myiasis. The presentation of pathology both in animals and in humans has been described in a previous paper, along with the characteristics of diffusion, the reproductive cycle of the larvae and the morphological and genetic findings.[16]

Rhinomyiasis is a rare disease in humans; therefore, literature data are still inadequate. Here, we report a rare case detected in Northern Italy, whereas previous cases were experienced in Sicily in 1965.[3] In addition, seven sporadic ophthalmomyiasis cases have so far been reported in Italy.[17–21]

Other cases of myiasis from returning tourists, especially men, have been registered worldwide—namely, in two English men returning from holidays in the Spanish Mediterranean coast[22] and Morocco[23] and in a Swedish man who had spent his holidays on a Greek Island.[7] Finally, also a woman was affected by the disease while on holidays in Miramas.[24]

The aim of this article is to analyse the correct methodology for the diagnosis and treatment of myiasis in human paranasal sinuses, as the infestation in the given location is likely to be misdiagnosed and not adequately treated owing to its rare manifestation, thus leading to potentially severe complications.

Owing to the small number of rhinomyiasis cases reported in the literature, no clear indications for its diagnosis and treatment are provided; therefore, therapies are difficult to standardise.

As for the diagnostic approach, a prompt myiasis diagnosis is crucial; it can exclusively be obtained through a fibre-optic endoscopy performed on patients presenting symptoms notwithstanding the standard antibiotic therapy. The endoscopic examination allows for differential diagnosis by detecting the presence of larvae and excluding other rhinosinusal pathologies. Once myiasis has been diagnosed, the execution of some preoperative evaluations including eye examination, maxillofacial CT and infectious disease assessment is suggested. The ophthalmological evaluation is fundamental for different reasons: first, because a

simultaneous infestation at the ocular level may be in progress; second, because the infestation may propagate from the nasal cavity to the ocular level through the lamina papyracea. Indeed, rhinomyiasis may cause orbital complications and local spread owing to the associated thrombophlebitis, affecting the bone and the adjacent cranial fossa.[15 25]

It would seem that, as underlined in all the most recent papers about rhinomyiasis, the execution of maxillofacial CT is always indicated. This should be urgently performed in addition to the endoscopic examination for a complete evaluation of paranasal sinuses and to exclude other rhinosinusal pathologies or a rare intracranial extension.[14 15] Furthermore, the CT study, performed as preoperative work-up for any endoscopic sinus surgery, is also mandatory in case of rhinomyiasis and allows the surgery to be directed exclusively towards the paranasal sinuses involved.

Rhinomyiasis can be idiopathic as in this case, but it is very important to make a differential diagnosis with underlying local pathologies that can favour the development of myiasis, such as sinonasal malignancies, atrophic rhinitis and neglected nasal foreign bodies.

As for therapeutic management, the few cases reported in the literature show no univocal approaches.

Some authors suggest, in addition to the systemic therapy, the execution of anterior rhinoscopy to remove the larvae and the administration of nasal decongestants; alternatively, the irrigation of the nasal cavity with 15% chloroform in oil or soaked with 1% ether, or turpentine oil is the suggested procedure.[1 26] In their paper,[22] Uriarte *et al* reported removing larvae by douching with normal saline, yet a further examination under general anaesthesia and CT revealed a breach of the cribriform plate with cerebrospinal fluid leak. Therefore, although today the endoscopic approach is effective, in the previous case rupture of the cribriform plate was a severe complication.

The first endoscopic removal of live maggots was reported in Britain, and it led to the resolution of disease without further complications.[27]

In our experience, prompt detection and mechanical removal of larvae through endoscopic approach under general anaesthesia have been an adequate and resolutive therapy, without any complications. Surgical removal, indeed, quickly resolved the symptoms; therefore, no anthelmintic therapy was necessary. We chose to perform functional endoscopic sinus surgery under general anaesthesia instead of removing the larvae under local anaesthesia to dominate the region of the osteomeatal complex, which appeared bilaterally oedematous and congested during the outpatient endoscopic examination. Furthermore, the maxillary sinus mucosa appeared bilaterally hyperplastic on CT; therefore, a bilateral antrostomy was performed to avoid the presence of remaining larvae in ethmoidal and maxillary sinuses.

Finally, on the basis of infectious disease examination and after the complete removal by surgery, no antiparasitic treatment was administered.

Learning points

- Even if rare, rhinomyiasis should be considered in travellers returning from holidays in tropical or rural Mediterranean regions showing rhinosinusitis-like symptoms, not regressing with usual therapies.
- Prompt diagnosis and exclusion of ocular, intracranial and rhinosinusal complications allow for simple and decisive surgical therapy.
- Endoscopic sinus surgery is the surgical gold standard.

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