ENT Updates

CASE REPORT

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Nasal Gout: A Rare Pathology

ABSTRACT

Gout is the most frequent cause of inflammatory arthritis. However, it may present atypical deposit sites for monosodium urate crystals, making its diagnosis difficult, particularly if there is no previous history of hyperuricemia. The aim of this study is to present a rare form of presentation of a common pathology such as gout with tophaceous gout involving the nasal dorsum as the first presentation. We report a 47-year-old male who presented at our clinic due to nasal deformity with progressive growth and a painful lesion located in the right nasal ridge. Due to the absence of other complaints or associated pathologies, and regarding CT evaluation which confirm a lesion on the right side of the nasal pyramid, with apparently well-defined limits, with an anteroposterior axis of approximately 10 mm, no diagnosis was possible without histologic findings. The patient underwent surgery to excise the lesion and the histological evaluation of the surgical specimen revealed a gouty tuft. Due to the aesthetic deformity, grafting was performed with cartilage and temporal fascia. An analytical evaluation confirmed hyperuricemia at the same time that another gouty lesion appeared on the left hand. He was referred to a rheumatology consultation and started medication with allopurinol. Thus this case draws our attention to the fact that common pathologies can have very atypical presentations.

Keywords: Nasal deformity, gout, hyperuricemia, chronic tophaceous gout

INTRODUCTION

There are several benign and malignant pathologies that cause deformity of the nasal dorsum or distortion of the nasal pyramid. These pathologies may include fibro-osseous tumors (osseous fibrous dysplasia, ossifying fibroma, and osteoma), granulomatous diseases (tuberculosis, histoplasmosis, blastomycosis, sarcoidosis, and histiocytosis), among others. Some storage diseases or metabolic diseases are included in this group, with some of these pathologies being extremely rare.[1]

Gout is a well-known abnormality in the metabolism of uric acid.[2] The disease consists of the accumulation and deposit of monosodium urate crystals. This occurs major in the peripheral joints and soft tissues presenting with the intermittent course of inflammatory joint manifestations with periods free of symptoms. And, although gout is most frequently seen in the extremity joints, such as the metatarsal-phalangeal, chronic tophaceous gout may develop as nodular lesions in all areas of the body including the head and neck region. Hardly, however, have there been cases reported that involved the nasal region.[3,4]

Diagnosis of gout can be clinically challenging, especially if it presents in an atypical location.[3,4]

We present a clinical case with a very rare presentation of a gouty tophus in the nasal dorsum.Main Points: +Gout is a rare cause of nasal deformity+Diagnosis can be difficult if no other signs of the disease are present. Main Points: +Gout is a rare cause of nasal deformity+Diagnosis can be difficult if no other signs of the disease are present. + Medical and surgical treatment is possible. Surgery can be propose for diagnostic reasons.

CASE PRESENTATION

Forty-seven-year-old male patient recurred to an otorhinolaryngology consultation complaining of a painful lesion at the nasal dorsum, with progressive growth and with about a year of evolution.

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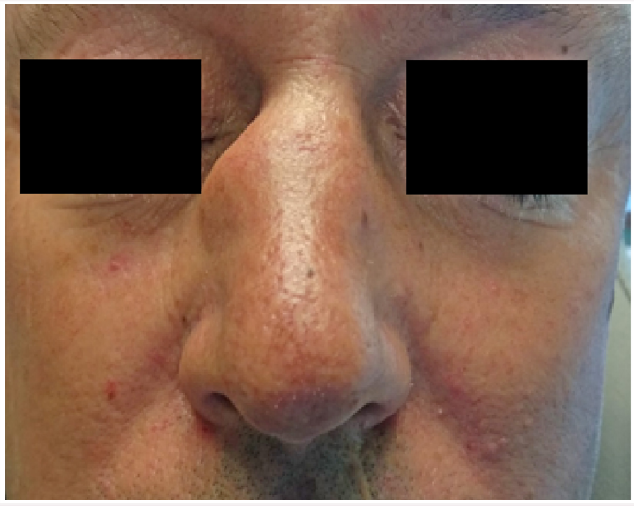

Figure 1. External nose deformity.

There were no complaints of nasal obstruction, rhinorrhea, sneezing or nasal itching, or smell changes. There was a history of nasal trauma due to alleged aggression about 10 years before. The patient denied other associated diseases or regular medication.

He brought an ultrasound report of that lesion that showed a lesion of about 12 mm in the dependence on subcutaneous cellular tissue with vascularization in the periphery. There were no other specifications.

On physical examination, he showed a firm and partial movable lesion on palpation that led to a deformity of the nasal dorsum on the right, with a diameter of approximately 1-1.5 cm. The lateral portion of the lesion, located over the nasal apophysis of the maxilla presented more elastic consistency, contrasting with the harder consistency of the remaining lesion. No inflammatory signs such as redness or external heat was seen (Figure 1).

In anterior nasoscopy and nasofibroscopy, nasal patency was maintained, with no signs of intranasal injury.

A computed tomography (CT) scan showed a lesion on the right side of the nasal pyramid, with apparently well-defined limits, with an anteroposterior axis of approximately 10 mm, height of 12.5 mm, and lateral axis of 6 mm. There was another component that extends anteriorly from the nasal flap to the most anterior portion of the nasal fossa, with asymmetry of the nasal bones, shorter on the right, in the region of this lesion. There was no bone invasion or infiltrative growth into the nasal cavities. Its density suggested extensive uptake of the contrast product that would be compatible with a vascular nature. Adjacent fat has normal permeability (Figure 2).

MAIN POINTS

- Gout is a rare cause of nasal deformity.
- Diagnosis can be difficult if no other signs of the disease are present.
- Medical and surgical treatment is possible. Surgery can be proposed for diagnostic reasons.

It was proposed for exeresis biopsy of the lesion under general anesthesia, for diagnosis purpose.

An intranasal transfixion and inter-cartilaginous incision approach was initially used. During the excision, a lesion with poor-defined limits and not encapsulated was identified, composed of a whitish lumpy material, with partial erosion of the right nasal bone, adherent to the skin, which was accidentally opened, although it was attempted an initial endonasal approach (Figure 3). The cosmetic defect of the nasal deformity created by the removal of the lesion was improved by filling the defect with temporal fascia and conchae cartilage fragments.

Two weeks after the surgery, no relevant cosmetic defect was seen (Figure 4).

Histological examination showed a cluster of basophilic, fibrolamellar acellular material, surrounded by histiocytes in the periphery, morphological features compatible with gouty tophi (Figure 5A and 5B).

In the postoperative evaluation, 2 weeks after surgery, the patient complained of a hardened lesion on the middle finger of the left hand, painful, and with external inflammatory signs (Figure 6).

Faced with this lesion and the histological findings, the patient was referred to a rheumatology consultation on suspicion of hyperuricemia already in the gout phase. He performed an analytical study that revealed a sedimentation velocity of 50 seconds and an uricemia of 8 mg/dL and started treatment with Allopurinol 300 mg and dietary care.

DISCUSSION

There are several pathologies that can lead to nasal deformity. Among these pathologies, there are deposit pathologies such as gout.

Gout is a well-known abnormality in the metabolism of uric acid. It is the most prevalent inflammatory rheumatic disease. Its prevalence is rising in the modern world, due to diet and lifestyle habits. The real prevalence of gout varies widely according to the population studied but ranges from <1% to 6.8%.[2-4]

The disease is stimulated by a diet rich in purine, which causes high levels of uric acid, which leads to the formation of monosodium urate crystals, which can be deposited in different organs. Hyperuricemia is defined as a serum urate level above the monosodium urate crystals saturation point (>6.8 mg/dL). At this point, the risk of crystallization increases. Our patient presented at a level of 8.0 mg/dL.[4-6]

Acute or chronic forms of presentation can be present. Acute forms appear as sudden, self-limiting attacks of arthritis (swelling, redness, pain, and warmth of a joint), while chronic forms result in the deposition of aggregates of crystals in and around the joints, causing deformity and progressive joint destruction.[6]

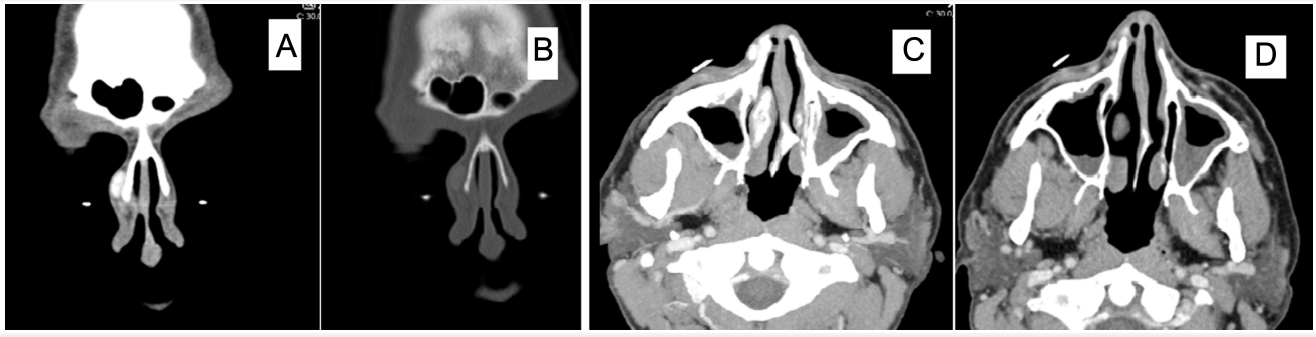

Figure 2. Computed tomography scan showed a lesion on the right side of the nasal pyramid, with apparently well-defined limits and with an anteroposterior axis of approximately 10 mm. A more anterior component extended anteriorly from the nasal flap to the most anterior portion of the nasal fossa, with asymmetry of the nasal bones, shorter on the right, in the region of this lesion (A and B coronal images and C and D axial images).

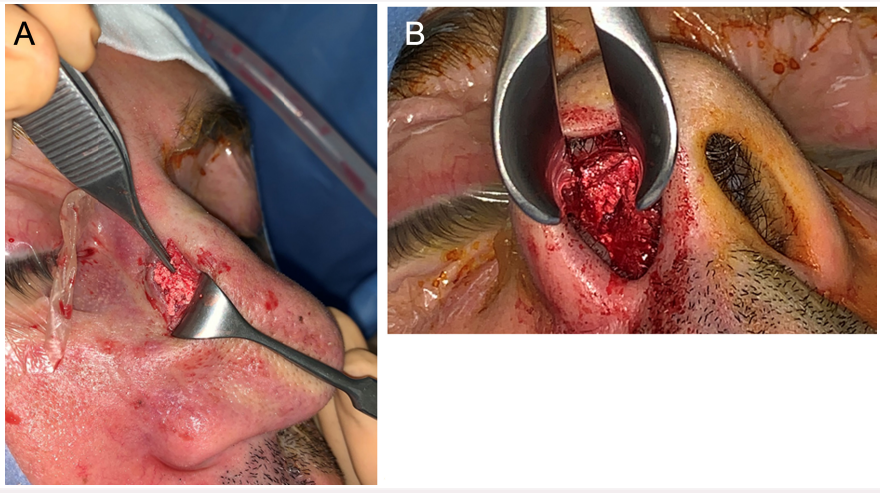

Figure 3. Intraoperative images with a whitish tissue of lumpy consistency, adherent to all deep tissues, including the right nasal bone, and adherent to the skin (A) external view and (B) intranasal view.

There are typical deposit sites, which include the peripheral joints, special from the lower extremities, particularly the first metatarsophalangeal joint, knees, ankles, and elbows. There are also atypical deposit locations, which include in the head and neck region the sclera, nose, or ears and heart valves, breast, abdominal striae, and gouty panniculitis.[5-7] It is very rare in the nose. As far as it was possible to find out, there are so far less than 10 cases described in the literature.

In our case, the patient had no history of gouty arthritis, and the nasal tophus was the initial presentation of gout. Most of the previously described cases had already identified hyperuricemia.

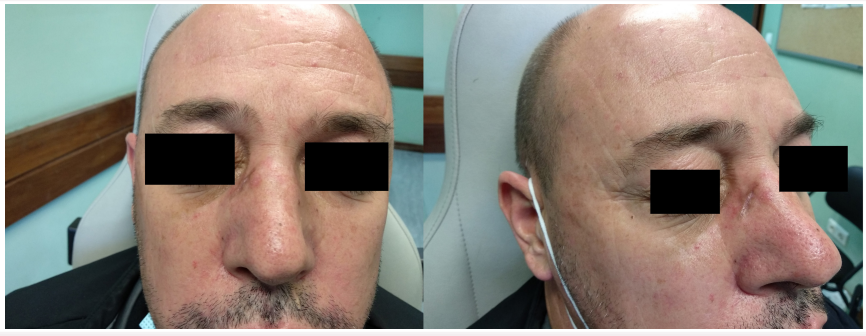

Figure 4. Postoperative frontal and lateral views.

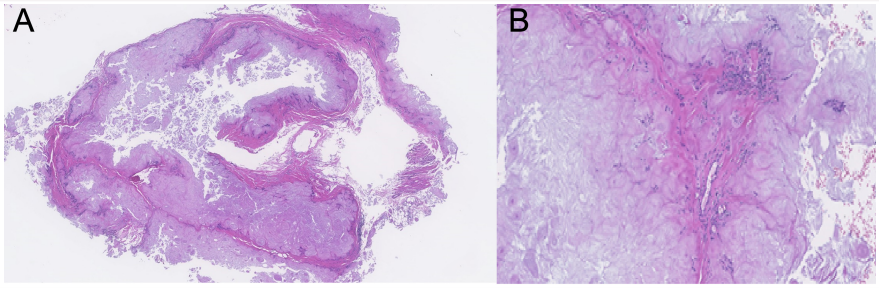

Figure 5. (A-B) Histologic examination showing clusters of basophilic, fibrolamellar acellular material, surrounded by histiocytes in the periphery.

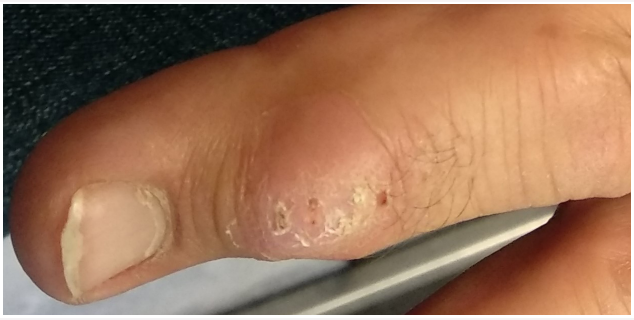

Figure 6. Left-hand lesion.

It is more frequent in males and usually begins between the ages of 40 and 60.[6]

It is a very painful and disabling disease. In this case, the patient complains of a painful lesion at the nasal dorsum which is innervated by the anterior ethmoidal nerve and the infraorbital nerve, and which can be damaged or irritated causing a painful sensation by tophus that can cause lytic changes.[5]

The description of this case serves to draw attention to a very frequent pathology that presents very rare atypical manifestations, especially as a primary presentation.

The diagnosis is made by the clinical suspicion correlating clinic findings, being easier if systemic hyperuricemia is identified, and imagological finding, but the final diagnosis is made under polarized microscopes with the appearance of strong negative birefringent needle-like monosodium urate crystals.[5]

Imaging could be helpful to establish a diagnosis, but neither CT nor magnetic resonance imaging has specific diagnostic findings. In the acute phase, only soft tissue swelling around the affected joint site may be demonstrated. The typical radiographic finding in chronic gout includes well-defined "punched-out" erosions with overhanging and sclerotic edges, radiodense soft tissue nodules (tophi) with or without calcifications and asymmetrical involvement.[3] New dual-energy CT can help differentiate urate and non-urate deposition with high sensitivity and specificity.[3]

Another diagnostic tool that must be considered is ultrasound scan (US) complemented with fine needle aspiration cytology. The ultrasound distinctive characteristics of gout tophi include hypoechoic to hyperechoic image, with an inhomogeneous appearance, frequently surrounded by a small anechoic rim, with no internal flow.[4]

In this case, the imaging could help as a diagnostic tool, and the definite diagnosis was made by the pathological examination of the surgical specimen.

In histologic findings, I can find the major structure of the tophus contains a nest of urate crystals surrounded by soft tissue and inflammatory cells, organized in 3 primary zones: a central core of urate crystals; a surrounding, high-density, cellular coronal zone; and an outer fibrovascular zone, containing macrophages and plasma cells, fewer mast cells, and scattered T and B lymphocytes.[4]

The treatment for persistent hyperuricemia is typically taken by lifestyle changes and medical therapy, with Probenecid, Allopurinol, Cochicine, and/or Febuxostat. The aim is to lower serum urate concentration to an optimal level in order to prevent acute gouty attacks or the formation of gouty tophi. When medical compliance is insufficient and there are joint limitations or structural damage or cosmetic reasons tophi lesions should be surgically removed.[3-5]

All other cases described in the literature received surgical treatment for their nasal gouty tophus. Their chief complaints, whether it was a cosmetic appeal, diplopia, or nasal obstruction, were improved postoperation. Reconstruction of the nasal bone defect can be done using bone or cartilage grafts, nasal prostheses, or metal plates.[4]

In this case, the reason for the surgery was mainly diagnostic complemented by cosmetic reasons. Both CT and surgery identified erosion of the right nasal bone. After the lesion was removed a reconstruction and grafting with cartilage fragments and temporal fascia was performed for cosmetic reasons. Overlying skin was not compromised so skin grafts were not required.

The authors present a rare form of gout with the involvement of the nasal pyramid. This case demonstrates the difficulty of diagnosing these atypical forms of gout, where neither the clinic nor imaging could help us in the diagnosis.

We are aware that the description of clinical cases is considered to be an invaluable form of scientific evidence, but it is from the collection of published cases that larger investigations can be carried out.

Informed Consent: Written informed consent was obtained from the patient who agreed to take part in the study.

Peer-review: Externally peer-reviewed.

Declaration of Interests: The author has no conflict of interest to declare.

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