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CASE REPORT

Bizarre parosteal osteochondromatous proliferation (Nora's lesion): A case with acute and painful presentation

Proliferación osteocondromática parosteal extraña (lesión de Nora): Un caso de presentación aguda y dolorosa

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Abstract

Bizarre parosteal osteochondromatous proliferation, also known as Nora's lesion, is relatively rare benign lesions that develop in small bones of hands and feet. In this study, one case of this disease in the proximal phalanx of the right index of a 42-year-old man has been reported. Physical exam and clinical characteristics, clinical, radio graphical and pathologic findings suggested the presence of this lesion. There was not any history of trauma and injury and in the exam, we find pain and swelling, mild local tenderness, extra osseous calcification in x-ray, and mass with high signal in T2 was observed. The mass was removed by surgery and observation showed no malignant manifestations. The patient did not show recurrence or complications after surgery.

Key words: Osteochondromatous, Nora's lesion, Trauma, Injury.

Resumen

La proliferación osteocondromatosa parosteal extraña, también conocida como lesión de Nora, es una lesión benigna relativamente rara que se desarrolla en los huesos pequeños de las manos y los pies. En este estudio se reporta un caso de esta enfermedad en la falange proximal del índice derecho de un hombre de 42 años. El examen físico y las características clínicas, los hallazgos clínicos, radiográficos y patológicos sugirieron la presencia de esta lesión. No había antecedentes de traumatismos y lesiones y en el examen se encuentra dolor e inflamación, leve hipersensibilidad local, calcificación extraósea en la radiografía y se observa masa con alta señal en T2. La masa fue extirpada mediante cirugía y la observación no mostró manifestaciones malignas. El paciente no presentó recidiva ni complicaciones tras la cirugía.

Palabras clave: Osteocondromatoso, Lesión de Nora, Trauma, Lesión.

Background

Bizarre parosteal osteochondromatous proliferation (BPOP) was reported by Nora et al. for first time[1]. It is also nomenclatures as Nora's lesion. This disease is a rare discomfort that occurs in small bones. However, large bone involvement has also been reported in some studies[2,3]. This complication is benign and in most cases, the rate of growth and relapse is low[4]; however, in rare cases, it's possible that it is mistakenly considered as malignancy due to the high rate of recurrence, rapid growth and histological findings and observations. In 20-50 percent of reported cases, this is a diagnosis of malignancy due to atypical diagnostic findings and histopathologic appearance[4]. In this study, a case of this disease with symptoms of rapid growth and pain in proximal phalanx of the right index finger in a 42-year-old man was reported. The diagnostic findings and treatment of this patient were also mentioned.

Case presentation

The patient was a 42-year-old man with a complaint of pain and swelling in the proximal phalanx of the right index finger (**Figure 1**). In the early studies, there was no history of trauma and ulcers in this phalanx. There was also no history of diabetes and metabolic disease in this patient. Local swelling and its related tenderness were observed in the proximal phalanx. On palpation, a mildly tender mass with a fairly hard consistency was touched on the finger. The neurovascular exam and movements of the finger were intact.

Investigations if relevant

In x-ray, soft tissue swelling accompanied by extra-bone calcification and the adjacent proximal lining was evident (**Figure 2**). Also in MRI, a mass with a high

signal at T2 was evident (**Figure 3**). Other radiographic studies including x-ray of chest and lung were normal. Laboratory tests were also in normal range. According to the rapid growing history of mass and painfulness of the lesion, an incisional sampling was considered for further investigation. The patient was followed up for one year and no recurrence or other complication has ever been established.

Figure 1: Anterior and posterior view, patient Rt hand.

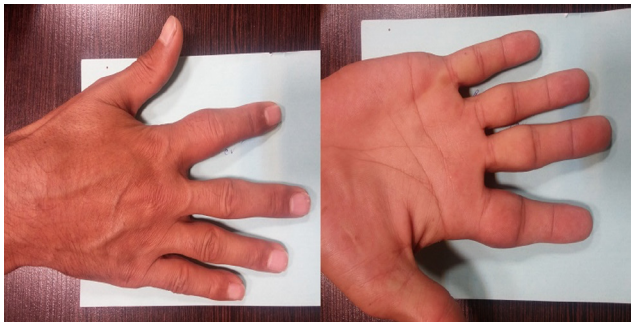
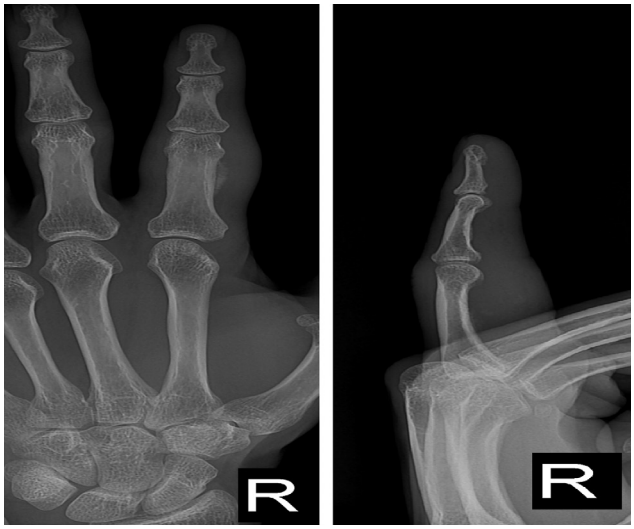


Figure 2: Plain X-ray ,patient Rt hand ,AP and Lateral.



Treatment

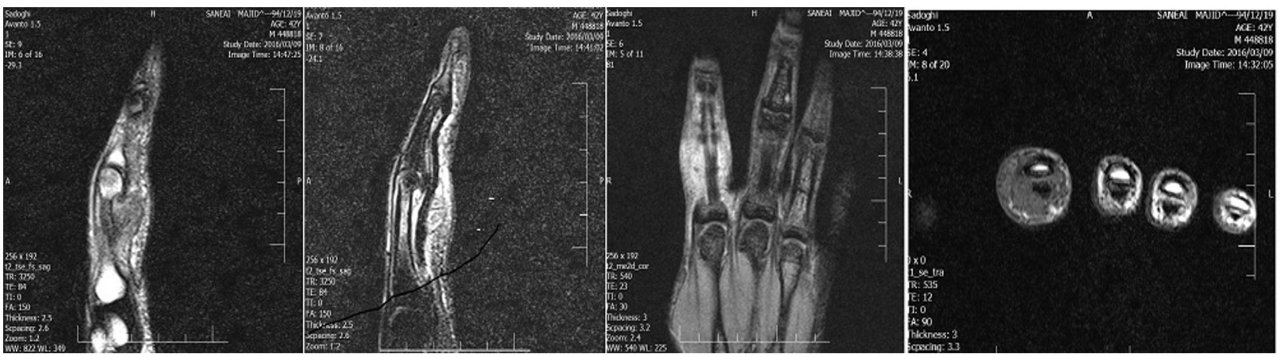
The surgery was planned by an oblique incision directly on the volar aspect of the proximal phalanx. We found a hard mass that was completely detachable from the proximal region. There was no atypical soft tissue around the mass. This mass had no clinical manifestations of malignancy and it was completely eliminated. So the lesion totally excised for microscopic examination. And the result was BPOP.

The patient was followed up for one year and no recurrence or other complication has ever been established

Discussion

BPOP or Nora is a rare condition that occurs in the form of mass in small bones. But, it is also reported in some other bones, such as the humerus, radius, etc[5,6]. The distribution of this type of disease is almost identical between men and women and there is no gender preference. The age distribution of this disease is greater in the range of 20-30 years old, although it can occur at different ages[2]. In our study, the patient's age was 42 years. The observed masses exhibit Histopathologically and radiographically distinctive features. In most cases, the history of trauma and infection has not been reported. The history of trauma has not also been mentioned in the reports of the patient in this study. In most cases, the lesion has been reported in the bones of the hand, and few cases have reported the disease in the bones of the leg[1]. The location of the lesion in the patient in this study, as in most cases reported in the hands, was in the proximal phalanx of the right index finger. The initial treatment of this disease is surgery, and since the lesion is benign, treatment is well-suited[7]. Removal of adjacent abnormal tissues is also recommended during surgery. Excessive resection is also not permitted, as it may result in loss of function especially in the hands and feet[3]. In our study, we also easily removed the mass and abnormal adjuvant tissue. In some cases, recurrence of the disease has been reported. These lesions tend

Figure 3: MRI ,T1 and T2 sequences and in different sections.



to be significantly reversible: relapse rates have been reported between 29-55% for a 2-year period, with almost half of these patients have been experienced the second recurrence. In terms of metastasis and mortality, no cases have ever been reported. The present study confirms this issue. The BPOP radiographic and microscopic features are reported in all reported cases as key points for the diagnosis and characterization of this disorder. These characteristics have led to confusion with other cases such as osteochondroma, osteosarcoma, chondrosarcoma and so on[8]. The best way to detect this lesion from osteochondroma is histopathologic findings. The distinction between BPOP and osteochondroma is important so that BPOP is more likely to require invasive surgery than osteochondroma and it has a higher rate of relapse[9]. In our study, the definitive diagnosis of this complication was determined by examination of histology and pathology. The first stage of BPOP appears as swelling of the soft tissue with or without quantitative calcification[3]. In this study, radiographic studies confirmed that there was a soft tissue swelling associated with extra osseous calcification, and the two-month disease history and painfulness of the lesion showed a possibility of mass malignancy. But during surgery, the result was a lack of malignancy in the mass. Gerald Gruber et al reported three cases of BPOP disease with symptoms of increased volume, pain without any history of trauma. In their report, the radiographic features of the calcified masses were involved in tissues around the bone. In MRI studies, there was also a mass with a high signal at T2. Yuichiro Matsui et al recorded calcification of the surrounding tissue and high signal in T2 in their case[10]. Typically, the mass has a low signal in T1 and a moderate to high signal in T2[9]. The radiographic characteristics of our studied case were similar to those of the above studies so that the mass with calcification and the high signal was observed Nora et al., Dhont et al., and Meneses et al. Reported local relapse of 51%, 29%, and 55%, respectively[2]. In this study, the patient did not show any recurrence or other complications after the one-year post-surgical follow-up. In some cases, the time between the first resection and recurrence was two months. Our patient did not show any recurrence in the follow-up. According to the observations of this study and comparison with other reported cases, due to the similarities between the disease and the osteochondroma (although it is rare)[8], it is difficult to detect this tumor precisely during the diagnosis process and it may be misdiagnosed. Painfulness of the lesion and quick clinical course are two important difference of our case with other reported cases.

- The use of hybrid diagnostic methods including radiographic, microscopic and histopathologic techniques for diagnosis of this disease is suggested
- Surgical procedure along with the removal of adjacent abnormal tissue is recommended as an effective treatment.

Follow-up of the patient for the possibility of recurrence and complication is recommended.

Competing interests

Author has declared that he have no competing interests.

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