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Intraosseous Lipoma of the Maxilla in a Patient with Henoch-Schönlein Purpura: A Case Report and Literature Review

Henoch Schönlein Purpuralı Hastada Maksiller İntraosseöz Lipom: Olgu Sunumu ve Literatür Taraması

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Abstract

Intraosseous lipomas are rare benign tumors of the bone that are mostly seen in the metaphysis of long bones and the calcaneus. Intraosseous lipomas are generally asymptomatic, and surgically removed when they are symptomatic or cause cosmetic concerns. Intraosseous lipomas of the maxillary bone have rarely been reported. In this report, we present a case of maxillary intraosseous lipoma in an 11-year-old girl with Henoch-Schönlein purpura-a rare case reported for the first time in the literature.

Keywords: Lipoma, intraosseous lipoma, maxillary bone, Henoch-Schönlein purpura

Öz

İntraosseöz lipomlar kemiğin nadir görülen benign tümörlerinden olup çoğunlukla uzun kemiklerin metafizinde ve kalkaneusta görülür. Genellikle asemptomatik olarak seyreden intraosseöz lipomlar, kozmetik kaygılar yaratıyorsa veya semptomatik ise opere edilirler. Literatürde maksilla yerleşimli intraosseöz lipom olgusu oldukça azdır. Bu yazıda Henoch Schönlein Purpuralı 11 yaşındaki kız hastada maksiller intraosseöz lipom olgusu sunulmuştur. Olgu, Henoch Shönlein Purpuralı hastada ilk maksilla yerleşimli intraosseöz lipom olgusudur.

Anahtar Sözcükler: Lipom, intraosseöz lipom, maksilla, Henoch-Schönlein purpura

INTRODUCTION

Intraosseous lipomas, first reported in 1880, are very rare and constitute 1 to 2.5% of all primary bone tumors.[1,2] Most intraosseous lipomas are asymptomatic and incidentally detected. They more frequently occur in men and in the fourth decade.[3] They are often located in the calcaneus and metaphyses of the long bones, in the lower extremity, but rarely seen in the cervical spine, ribs, pelvis, upper extremity, skull, mandible, and maxilla.[4,5] Here, we present a first case of an intraosseous lipoma of the maxilla in a patient with Henoch-Shönlein purpura (HSP) and our review of the literature.

CASE REPORT

An 11-year-old girl was admitted to our clinic for a swelling on the right side of her nose that has been slowly growing for almost 5 years. The mass measured approximately 2×1 cm; it was firm and immobilized and located 2 cm below the right medial canthus, expanding the skin (Figure 1). She was diagnosed with HSP at the age of five, when she presented with abdominal pain, arthralgia, and purpuric skin rash. The patient underwent computed tomography (CT-Somatom HiQ; Siemens, Erlangen, Germany) of the maxillofacial region. The CT scan showed a hypoattenuated mass of approximately 5x10 mm in diameter to the anterior wall of the right maxillary sinus at the level of the nasomaxillary junction. Further, the patient underwent magnetic resonance imaging (MRI-Magnetom SP 42 E, Siemens, Erlangen, Germany) and examined with a 1.5-T superconductive imaging unit. T1-weighted images demonstrated a high-signal intensity lesion with signal loss on fat suppressed images.

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A well-contoured mass was observed to cause remodeling adjacent bone cortex and mild thinning without destruction. The mass was homogeneous with no differentiated tissue components. After contrast enhancement following magnetic resonance imaging was performed, the mass was diagnosed as benign intraosseous lipoma (Figure 2). The subperiosteal mass was excised with a right maxillary gingivobuccal incision in supine position under general anesthesia. The section was primarily sutured after bleeding control. The mass was preserved in 10% formol solution for histopathological examination. Atrophic bone lamella surrounded by mature fat tissue and focal calcification were detected on histopathological examination of the mass. The absence of lesion, necrosis or mitosis in the bone marrow tissue, led to a diagnosis of benign intraosseous lipoma (Figure 3). The patient was discharged on the first postoperative day without any complications. During the follow-up period the patient reported complaints of paresthesia along the infraorbital nerve at the first month. It regressed at the sixth postoperative month without evidence of recurrence. (Figure 1).

Written informed consent was obtained from the first degree relative of the patient.

DISCUSSION

Intraosseous lipomas are usually observed in individuals aged 30–60 years, with the highest prevalence in the fourth decade. The youngest and oldest cases reported to date are those of a four-year-old and an 84-year-old, respectively. Lipomas are more predominant among males.[1-3]

The etiology of intraosseous lipomas is not completely understood. Some authors advocate that it is a benign tumor

Figure 1.a,b. Intraosseous lipomas with minimal expansion of the skin approximately 2 cm below the medial canthus on the right lateral side of the nose. (a) Preoperative view (b) View of patient at postoperative month 1

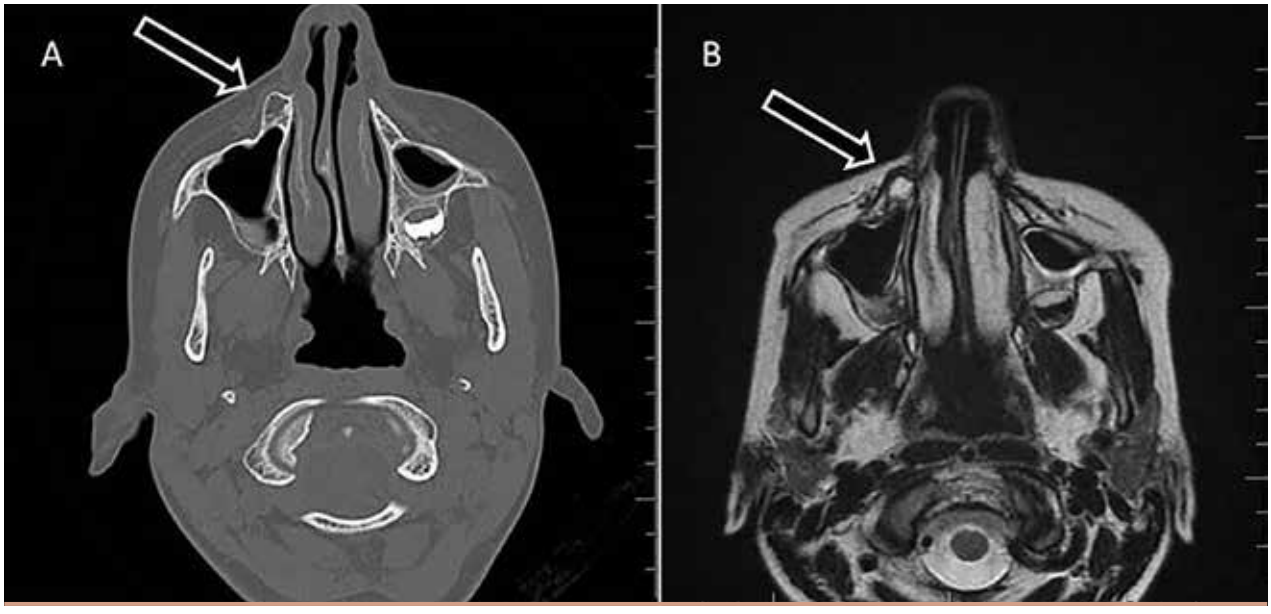

Figure 2.a,b. Intramedullary intraosseous lipoma in the right nasomaxillary junction. (a) Computed tomographic image of the mass with fat density that caused adjacent cortex remodeling and thinning. (b) Hyperintense appearance of the same mass in T1-weighted sequence of MRI

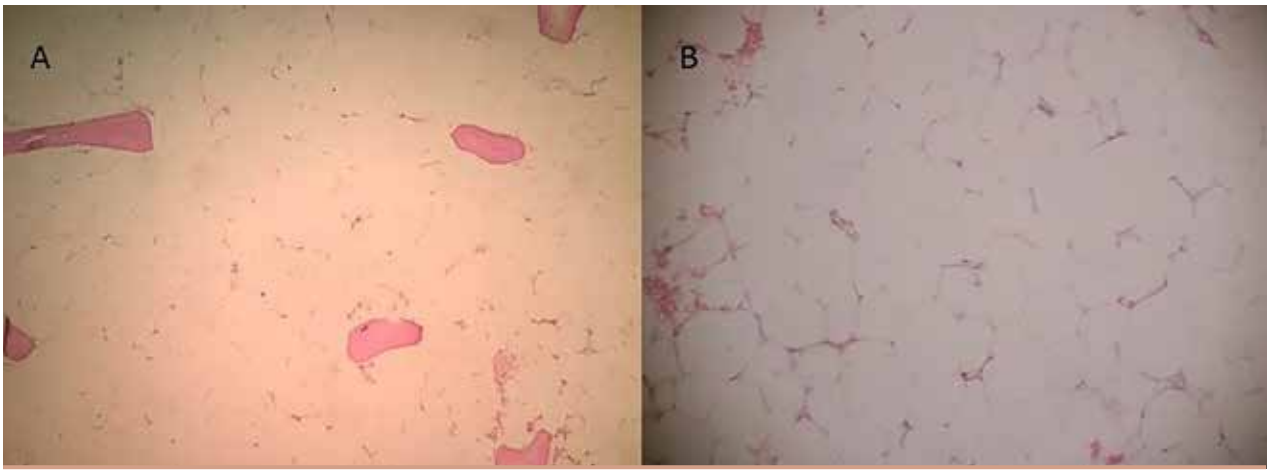

Figure 3.a,b. Mature fat tissue and focal calcification areas surrounding the atrophic bone lamellae with light photomicroscopy. Hematoxylin-eosin staining: 10× (a) and 100× (b)

of medullary fat tissue, whereas many authors believe that it occurs secondary to infarction, infection, or post-traumatic bone reaction. [4,6-8] Yet, none of these theories clearly explain the etiology of intraosseous lipomas.

Milgram classified the histopathological findings of intraosseous lipomas in three groups.[2] In stage one, there are mature lipocytes between fine bone tubercles. Stage two is similar to stage one, but there are infarct and calcification areas between rigid tubercles due to the expansion of fat cells. In stage three, fat necrosis, calcified fat, cyst formation, and reactive peripheral and central new bone formation are seen with enlarged infarct areas. The presence of mature fat cells and focal calcification between the atrophic bone lamellae indicates that the lesion is in the second stage.

Plain radiographic characteristics of intraosseous lipomas vary according to histopathological staging, which in 1988 Milgram has classified based on his findings.[9] Although detailed radiological classification has been described, the findings are non-specific and insufficient for diagnosis and differential diagnosis. Plain radiographic characteristics of intraosseous lipomas may mimic many lesions such as fibroma, aneurysmal bone cysts, fibrous dysplasia, osteoblastomas, chondrosarcomas, giant cell tumors, bone infarcts, chondroid tumors, rarely osteomyelitis, metastasis, eosinophilic granuloma. Therefore, advanced imaging methods such as computed tomography and magnetic resonance imaging are needed for differential diagnosis.[6,7] Campell et al. have reported that histopathological sampling is non-essential for diagnosis and that definitive diagnosis is possible with computed tomography and magnetic resonance imaging.[1] Intraosseous lipomas show as hypoattenuated lesions in computed tomography examination. Studies using magnetic resonance imaging indicate that lipomas have high-signal intensity characteristics on T1- and T2-weighted sequences, without contrast enhancement after contrast administration. Lipomas show signal loss on fat-suppressed sequences.[10]

In addition to histopathological classification, intraosseous lipomas are classified into three groups according to the tissue from which they originate: bone marrow-derived mass, also called intramedullary lipoma, is mostly seen in long bones; parosteal lipoma originating from the periosteum and leading to cortical bone reaction; and soft tissue lipoma, which is similar to parosteal lipoma and originates from soft tissue adjacent to the bone and causes compression or penetration.[5] The case presented in this article is intramedullary lipoma originating from the bone marrow, expanding the bone cortex.

While most intraosseous lipomas are asymptomatic, symptomatic ones present in different forms depending on the size and localization. Intraosseous lipomas located in the lower extremities clinically present with edema and pain because of microfractures caused by minor trauma.[1] Intraosseous lipomas located in the maxillary bone can be associated with pain and dental and gingival problems depending on the location of the lesion. The case presented in this article was asymptomatic, and the patient was admitted with a growing right mass.

Treatment of intraosseous lipoma is conservative. Surgery is performed in case of pathological fracture or cosmetic concern, for mass with risks of malignancy, and for symptomatic cases such as pain and paresthesia. The procedure involves curettage and reconstruction with backfilling cancellous bone graft.[1-3] In our case, surgery was performed because of the rare location of the mass, the young age and the cosmetic concerns of the patient. The mass expanding the bone cortex was excised, but bone graft was not applied during surgery since no bone defects and stressed areas were observed. The rate of recurrence and malignant transformation in intraosseous lipomas are very low. In the literature, recurrence is reported in only two cases and malignant transformation in five cases.[11-14] No recurrence was detected in the presented case after six months of follow-up.

Henon-Shönlein purpura is the most common type of vasculitis seen in children. The etiology of the condition, which affects small blood vessels and causes damage, is not completely known. This frequently self-limiting condition usually occurs at the age of five or six. It affects the skin, gas-

trointestinal tract, joints, kidneys, and rarely other organs.[15] There are no clinical data indicating a relationship between Henon–Shönlein purpura and intraosseous lipoma. Considering the theories related to its etiology, in the presented case intraosseous lipomas can be deemed to have developed secondary to a bone reaction with the first vasculitis attack. The fact that the mass was observed approximately one year after the diagnosis of Henon–Shönlein purpura support this theory.

CONCLUSION

Intraosseous lipomas are benign bone tumors with a low risk of malignancy and recurrence. Histopathologic sampling is not necessary for diagnosis, as computed tomography and magnetic resonance imaging may be sufficient. The treatment is conservative and if the mass causes cosmetic concerns or is symptomatic, it may be surgically removed.

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