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CASE REPORT

Nasal angioma with osseous metaplasia

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ABSTRACT

Nasal angiomas are rare. We report a case showing osseous metaplasia and discuss pathogenesis hypotheses. A 41-year-old woman presented with a right lateronasal firm, immobile mass, and interfering with glass wearing. The computed tomography scan imaging was suggestive of chondroma while the magnetic resonance imaging showed on T1-weighted images nodule isosignal, on T2-weighted images hypersignal, and intense enhancement after contrast substance injection. The lesion was surgically resected. Histological examination revealed a 0.8 cm angioma with multifocal osseous metaplasia. The diagnosis of nasal angiomas with extensive osseous metaplasia is difficult requiring microscopic examination. Conservative surgery is the treatment of choice even at an early stage due to the limited effectiveness of embolization or drugs on the osseous component.

Key words: Angioma, benign vascular lesion, diagnostic, nose osseous metaplasia, vascular malformation

INTRODUCTION

Nasal angiomas are rare.[1] They may rise from the nasal bone or from soft tissues. We report a case of nasal angioma peculiar by the presence of osseous metaplasia and discuss pathogenesis hypotheses.

CASE REPORT

A 41-year-old woman presented with a right lateronasal firm, nonmobile mass, progressively developing after trauma for 15 years. The lesion was interfering with the wearing of the glasses. Clinical examination showed no oculomotor symptoms, intranasal lesion, internal fistula, or skin change. There was no relevant medical history apart from viral hepatitis (childhood), eczema (date unknown), menorrhagia since the age of 23 years (treated for 28 months by monophasic estroprogestative drugs containing 0.02 mg ethinyl estradiol and 0.1 mg levonorgestrel, treatment begun after surgical resection of a 2 cm vaginal endometriosis nodule) and occasional epistaxis, and bleeding gums. The familial medical history revealed a skull tumor in the father (type, treatment: unknown). The arterial blood pressure and glycemia were normal; the bone mass index was 27.1 and the blood group type A. The blood tests were fluctuant showing preoperatively, low hemoglobin (11.9 g/L, normal: 12–15) and hematocrit (35.2%, normal: 38–48), normal thrombocytes (220,000/mm^3), normal kaolin clotting time ratio (1.11, normal < 1.20), increased activated cephalin time ratio (1.21, normal < 1.20), and increased factors I/fibrinogen (4.06, normal 2–4 g/L), IX and XII (126 and 153, respectively, normal: 70–120 for both). On computed tomography scan, there was a 0.6 cm expansile, osteolytic lesion of the right nasal wing. The diagnosis of chondroma was mentioned. On the subsequent magnetic resonance imaging, the nodule, at contact to the right nasal bone, measured 0.7 cm and showed on T1-weighted image isointense signal, on T2-weighted image hyperintense signal and after intravenous gadolinium injection, intense contrast enhancement [Figure 1]. The bulk of the nodule was in the

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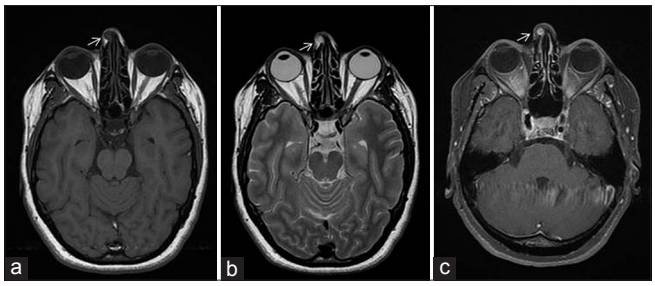

Figure 1: The nasal lesion (arrows) showed on T1-weighted image at magnetic resonance imaging isosignal, on T2-weighted image hypersignal, and intense contrast enhancement after gadolinium injection (a, b and c, respectively). The brain and nose sections were not completely overlapping on the three images and a zone of artefact appeared on image c possibly as related to the procedure/image processing/movement

subcutaneous soft tissues, adherent to the nasal bone and without change to the overlying skin or intranasal bulging. The surgical resection of the nodule was uneventful. Three months after operation, the patient was free of recurrence and showed increased activated cephalin time ratio (1.21 and increased factors I/fibrinogen, VII, IX and XII (4.57, 122, 126, and 153, respectively).

The resected specimen measured 0.8 cm. After decalcification, the microscopical analysis (performed on several serial- and multi-step-sections of the entire specimen) revealed a rounded lesion in part limited by a rim of dense connective tissue and with an appended fragment of normal adipose tissue [Figure 2]. The nodule consisted mainly of an aggregation of abnormal, cavernous, irregular-shaped and different-sized, large blood vessels, and cavities. There were no arterial-type vessels with identifiable elastic laminas or calcifications within the vessel walls. The vessels' wall showed irregular thickness, with a rim of smooth-muscle-actin positive cells and intraluminal malformative excrescencies (tuft-like). Several vessels were noted at the periphery of the lesion. Bone fragments composed mainly of woven bone were observed within the entire nodule, both in the vessels' wall and in the scarce intervascular connective tissue. These osseous trabeculae showed numerous osteocytes and focally, rimming osteoblasts. There was no inflammation, thrombi, or organized hematoma.

DISCUSSION

Here, we report a rare type of vascular lesion, peculiar by the presence of extensive osseous metaplasia. The morphology of the vascular spaces suggested an angioma. On the other hand, the presence of the intraluminal tuft-like excrescencies (rarely encountered in hemangiomas) indicated the possibility of a vascular malformation.[2] Interestingly, the vessels' wall also showed a rim of smooth-muscle cells of varied thickness, a feature consistent with that seen in malformative shunt

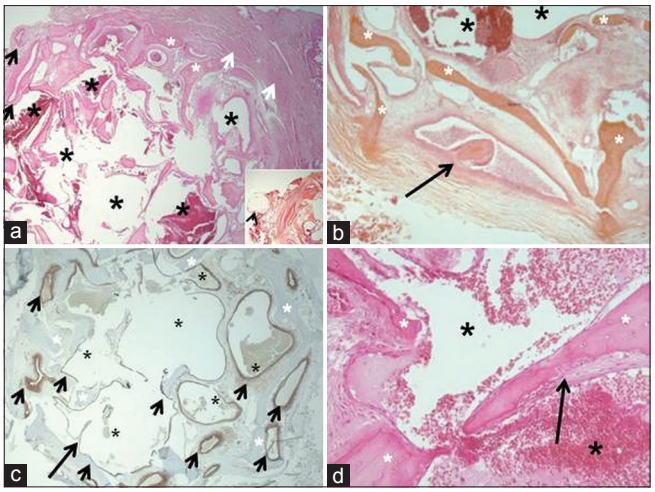

Figure 2: On microscopical examination, the lesion consisted in large vessels and vascular cavities, and disperse bone fragments (a-d: Black asterisk/vascular cavity; black short arrows/large blood vessels, white asterisks/osseous metaplasia). Several vessels or vascular cavities showed tuft-like wall projections/excrescencies (b and c: Black long arrow). The nodule was limited focally by a connective tissue rim and a fragment of adipose tissue appended (a: White arrows, a inset: Arrowhead for the adipose tissue). The vessel wall was of varied thickness (b: Black short arrows/vascular wall of varied thickness, with smooth-muscle-actin positive layer). Osseous metaplasia was multifocal, disperse in the entire nodule, in the vessel wall and in the sparse connective tissue the vascular structures (a-d: White asterisks). (a, b, and d) H and E stain, b immunohistochemistry for smooth-muscle-actin; original magnification ×2.5 (a, a inset, c); ×5 (b); ×10 (d)

vessels. In the light of sex-hormone treatment, the patient had for menorrhagia and endometriosis, we could not eliminate the impact of these drugs on vascular smooth-muscle hyperplasia, similar to that reported in endometrial arterioles during oral contraceptive treatments.[3,4]

The diagnosis of nasal angioma at an early stage is difficult. The clinical examination may suggest an osteoma. This hypothesis was not confirmed by the computed tomography scan that showed an osteolytic lesion and by the magnetic resonance imaging that showed an intense enhancement after contrast substance injection. Once the diagnosis of angioma made on microscopical analysis of the resected specimen, the precise origin, whether primitive osseous or subcutaneous remained difficult to precise although an extensive examination on serial- and multi-step-sections and despite the small size of the lesion. The amount of bone tissue within the angioma, exceeding that of <2 mm thick nasal bone along with the random disposition of osseous fragments of woven bone-type and with the subcutaneous prominence and predominance of the nodule (and not intranasally) rather suggested a soft tissue angioma than an intraosseous angioma with exuberant osteoformation. Although bone formation in vascular lesions is very rare, this is reported even at distance from bones.[1,5,6] The large size of the vascular spaces and vessels along with the limited extension of

the intranodular connective tissue (as compared to the vascular component) and without significant inflammation allowed us to eliminate the hypothesis of a posttraumatic hyperplastic granulation tissue reaction. However, an association of ancient reparative lesions cannot completely ruled out. The hypothesis of ossified phleboliths/thrombi was excluded due to the lack of intravascular thrombi at microscopy although a background of abnormal blood coagulation. In the present case, the association between the angioma and the serum increase of factors I, IX and XII, among which, factor IX reported as risk factor for venous thromboembolism,[7] was probably incidental. To our knowledge, one case of ulcerated dermal angioma is reported in association with hyperfibrinogenemia, this finding being rather related to the patient's underlying Hodgkin's lymphoma.[8] In the case we report, of clinical interest would be the history of sex-hormone drug intake, known to possibly favor a pro-thrombotic background.

CONCLUSION

The diagnosis of nasal angioma is difficult, requiring microscopic examination. Conservative surgery can be performed at an early stage. Although possibly of limited effectiveness on the intraangioma extensive osseous metaplasia, embolization or medical treatments including with Cox-2 inhibitors[9,10] could be considered as an option.

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Conflicts of interest

There are no conflicts of interest.

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