CASE REPORT

Intraosseous cavernous haemangioma of the inferior turbinate

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SUMMARY

Intraosseous cavernous haemangiomas usually occur in the vertebral and calvarial bones, and account for <1% of primary bone tumours. Nasal cavity intraosseous cavernous haemangiomas are extremely rare. We present a case of a 47-year-old woman with an incidental left inferior turbinate mass noted on CT scan for an unrelated pathology. She had no rhinological symptoms other than a 2-year history of left-sided nasal obstruction. Preoperative imaging failed to determine the aetiology of the mass. The lesion, contiguous with the left inferior turbinate, was successfully resected via an endoscopic approach. Histopathological examination of the resected specimen demonstrated a left inferior turbinate intraosseous cavernous haemangioma.

BACKGROUND

The nasal cavity can harbour a wide variety of tumours, with haemangiomas arising from the soft tissues accounting for approximately 20% of benign nasal cavity tumours.¹ Intraosseous haemangiomas represent 0.7–1% of all bone tumours and primarily originate from the spine or skull.¹ Intraosseous cavernous haemangiomas of the nasal cavity are extremely rare with only four cases arising from the inferior turbinate reported in the literature.^{2–5}

We present a case of a patient with a nasal mass diagnosed postoperatively as an intraosseous cavernous haemangioma arising from the left inferior turbinate. The diagnostic challenge and management of this rare lesion is discussed.

CASE PRESENTATION

A 47-year-old woman was referred to our otolaryngology department from the regional Neurosurgical Centre following surgical management of a frontal lobe meningioma. Preoperative CT scan identified an incidental left nasal cavity mass (figure 1). She had a 2-year history of left-sided nasal obstruction in the absence of any other symptoms such as facial pain or epistaxis. There was no prior history of nasal packing or trauma. Flexible nasendoscopy revealed a minimally displaced septum and a firm, smooth, obstructing mass arising from the left inferior turbinate (figure 2). The mucosa was intact and non-hypervascularised. No other abnormality was found on examination of the head and neck.

INVESTIGATIONS

Serum haematological and biochemical tests were within reference ranges. CT scan and MRI were already performed by the referring hospital (prior

to surgical management of a large anterior cranial fossa meningioma) and reviewed in our multidisciplinary team meeting. CT scan demonstrated a 15 mm ground glass bony lesion (figure 2) involving the left inferior turbinate (possibly an exostosis) and a prominent right middle turbinate concha bullosa. There was no obstruction of sinus drainage pathways. The surrounding tissues were normal. Subsequent MRI further delineated the mass (figures 3 and 4) and gadolinium enhancement revealed the lesion to be diffusely avidly enhancing.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis at this stage included fibrous dysplasia, however, there are only two case reports in the literature of mono-ostotic disease involving the inferior turbinate.^{6 7} Furthermore, CT scan showed thickening of the inner table of the frontal bone, not involving the entire cortical thickness, as would be expected with fibrous dysplasia. It was thought that the lesion may represent an ossifying fibroma, although this was unlikely given such lesions are usually more aggressive and the surrounding tissues were normal in this case.

As the diagnosis remained in doubt, a whole body single-photon emission CT (SPECT) scan was performed as fibrous dysplasia shows avid uptake and may demonstrate other hotspots. This scan was inconclusive, demonstrating only faint increased uptake relating to the left inferior turbinate lesion. Mono-ostotic fibrous dysplasia remained within the differential diagnosis.

TREATMENT

In the absence of a diagnosis, an endoscopic resection of the mass contiguous with the left inferior turbinate was performed under general anaesthetic (figure 5). There was no significant intraoperative blood loss. The patient made an uneventful recovery and was discharged on postoperative day 1.

OUTCOME AND FOLLOW-UP

Macroscopic examination of the surgical specimen revealed a 45×20×10 mm mucosal covered bony lesion. Microscopically, the lesion consisted of dilated vascular spaces interwoven between the bony architecture of the left inferior turbinate, without cytological atypia. Focal intraluminal thrombosis was seen. The surface epithelium was respiratory in nature with no dysplasia identified.

The histopathological examination revealed the lesion to be an intraosseous cavernous haemangioma arising from the left inferior turbinate. The

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Figure 1 Axial CT scan with bony left inferior turbinate lesion evident.

patient was asymptomatic with no evidence of recurrence at 1-year follow-up.

DISCUSSION

Haemangiomas arising from the soft tissues of the head and neck region are relatively common, however, nasal haemangiomas are rare.³ The most common sites for intranasal haemangiomas are the nasal septum, the lateral wall and the vestibule.⁸ Intraosseous cavernous haemangiomas account for <1% of primary bone tumours, and usually arise in the vertebral and calvarial bones.⁹ They are most common in women in their fourth and fifth decades.¹⁰ Intraosseous cavernous haemangiomas of the inferior turbinate are extremely rare with only four cases reported in the literature. Haemangiomas are classified

Figure 3 T2-weighted sagittal MRI demonstrating bony lesion involving the left inferior turbinate (frontal lobe meningioma also present).

into three subtypes, depending on their histopathological characteristics: capillary, cavernous and mixed. Cavernous haemangiomas have large blood-filled spaces lined with flattened endothelium, which often has papillae projecting into the lumen.

Local trauma has been proposed as an aetiological factor for intraosseous cavernous haemangiomas, however, a causal relationship is uncertain.³ The patient in this case did not report any history of nasal or head trauma.

Figure 2 Endoscopic view of left nasal cavity lesion contiguous with the inferior turbinate.

Figure 4 T1-weighted coronal MRI.

Figure 5 Endoscopic view of left nasal cavity postresection of the inferior turbinate and lesion.

Diagnosis of intraosseous haemangiomas can be challenging preoperatively. They are often asymptomatic lesions found incidentally on radiological imaging for other indications. Symptomatic lesions may present with nasal obstruction due to the mass effect or, rarely, recurrent epistaxis. Epistaxis may occur less commonly than in other mucosal haemangiomas because the lesion is covered with normal mucosa.⁹ Pain is not usually a feature. Our patient did not report any specific symptoms other than nasal obstruction, which is in keeping with other reports in the literature.^{2–4}

Intraosseous cavernous haemangiomas may demonstrate a characteristic appearance on radiological imaging. CT scan is helpful in establishing a diagnosis and sometimes demonstrates a well-circumscribed lesion with a characteristic ‘honeycomb’ appearance of the cavernous spaces within the lesion and a ‘sunburst’ pattern of trabeculations.² Phleboliths may be found within cavernous haemangiomas. MRI is useful to further delineate the lesion, determine the extent of impingement on surrounding tissues and to exclude intracranial, intraorbital or perineural extension. Despite their radiological characteristics, diagnosis of nasal intraosseous haemangiomas is difficult due to the absence of specific symptoms and the lack of signs to suggest a vascular lesion, for example, bluish discolouration, spontaneous haemorrhage and pulsation.³

Therapeutic options for the treatment of nasal cavity intraosseous haemangiomas include surgery, radiotherapy, sclerotherapy and embolisation.^{3 8 11} Haemangiomas have been shown to be radiosensitive, however, the risk of regional growth arrest and malignancy render radiotherapy unfavourable.² Therefore, radiotherapy is reserved for unresectable lesions.¹² Embolisation and sclerotherapy have been used to treat these lesions but these are palliative procedures.^{3 13} Studies have shown preoperative embolisation to reduce this risk of intraoperative bleeding in perioral haemangiomas.¹⁴ However, its role remains controversial because a definite vascular supply cannot be identified in the majority of cases, and the benefits are small compared to the potential risks of embolisation.^{10 15} Furthermore, our case of an inferior turbinate cavernous haemangioma and those reported in the literature have not been associated with any significant bleeding.^{2 3 9 10} Therefore, vascular intervention is not required if the lesion can be removed en bloc with adequate resection margins.^{15 16}

The treatment of choice for nasal intraosseous haemangiomas is complete surgical resection with sufficient margins. Our case, based on CT scan and MRI, was deemed amenable to endoscopic resection, which is less invasive than the Caldwell-Luc or midfacial degloving procedures adopted by two previous case reports in the literature.^{2 4}

In summary, intraosseous cavernous haemangiomas are rare and difficult entities to diagnose. CT scan aided by MRI is useful for preoperative evaluation. En bloc resection represents the treatment of choice for definitive management and for the prevention of recurrence.

Learning points

- ▶ Intraosseous haemangiomas should be included in the differential diagnosis of a bony nasal cavity mass.
- ▶ Differential includes fibrous dysplasia, ossifying fibroma, osteoma, osteogenic sarcoma and multiple myeloma.
- ▶ Nasal intraosseous cavernous haemangiomas often display characteristic radiological appearances, but this does not always lead to a definitive diagnosis. Therefore, surgery has a key role in the diagnosis and management of these lesions.
- ▶ Haemangiomas arising from the inferior turbinate are amenable to endoscopic resection with minimal blood loss.

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