CASE REPORT

Benign sinonasal capillary haemangioma

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SUMMARY

Haemangiomas are benign fibrovascular tumours relatively that are common in the head and neck, where 60% of them occur. Among the various categories of haemangiomas, lobular capillary haemangiomas (LCH) occur frequently on the skin, lips, buccal mucosa, tongue and gingiva. However, they are vanishingly rare in the paranasal sinuses. The imaging features of LCH are non-specific. The histological characteristics of LCH can also make diagnosis difficult as sometimes resemble highly vascular malignant tumours. This leads to the false preoperative diagnosis of suspected malignancy in many cases, which places the patient under unnecessary distress and anxiety. We present a case of LCH appearing as a suspicious nasal lesion of the lateral nasal wall with unilateral nasal obstruction, necrotic centre and epistaxis. The tumour was excised endoscopically in one operation including endoscopic medial maxillectomy. Further education about this lesion is deemed important as preoperative embolisation may be needed for a safe operation.

BACKGROUND

Haemangiomas are benign fibrovascular tumours first described in the 19th century. They are relatively common in the head and neck, where 60% of them occur.¹ Lobular capillary haemangiomas (LCH) are described by Mills *et al*² as a particular subset of vascular lesions of the skin and mucosal membranes with a diagnostic 'lobular arrangement of capillaries at its base'. They commonly present as a bleeding mass of sudden onset and are also known as pyogenic granulomas or human botryomycosis.³ LCH occur frequently on the skin, lips, buccal mucosa, tongue and gingiva. However, they are vanishingly rare in the paranasal sinuses.⁴ The imaging features of nasal haemangiomas are non-specific. Histologically, LCH can sometimes resemble either highly vascular malignant tumours, due to their pattern of endothelial proliferation and brisk mitosis, or granulation tissue, due to surface ulcerations and secondary inflammation. These characteristics of LCH also make histological diagnosis difficult. This leads to a false preoperative diagnosis in many cases, usually of suspected malignancy, which places the patient under distress and anxiety.⁵ Moreover, removal of these tumours without previous recognition may be technically difficult due to the increased vascularity and the risk of bleeding. A preoperative embolisation may be indicated.

CASE PRESENTATION

A 68-year-old woman presented to the ear, nose and throat department after referral from her general practitioner with a 10-week history of right-sided

nasal obstruction, associated with epistaxis from the same side. There was no history of prior surgery, trauma or epistaxis preceding the nasal obstruction. She initially had some facial pain, but this resolved after treatment with amoxicillin and a nasal steroid spray. She had no nasal discharge or known allergies, nor did she smoke or drink alcohol. She had, however, recently undergone excision of a lentigo maligna lesion from her left cheek.

On flexible nasendoscopy, a right-sided mass was seen, thought to be originating from the inferior turbinate or the maxillary sinus (figure 1). The mass was not actively bleeding; however, its size prevented further advancement of the endoscope on that side. No abnormalities were detected on the left side or in the postnasal space, larynx or pharynx.

INVESTIGATIONS

She was urgently investigated with CT and MRI scans of her sinuses, under the 2-week wait guidelines for suspected cancer, while also being prescribed Augmentin for 3 weeks, in case the mass was of infective origin.

The CT scan was reported as showing a rounded soft tissue mass extending from the floor of the right nasal cavity to the inferior turbinate, with destruction of the medial wall, extension into the posterior choanae and prolapse into the maxillary sinus.

DIFFERENTIAL DIAGNOSIS

The suspected diagnosis was one of inverted papilloma or squamous cell carcinoma, and so she was listed for urgent surgery. The differential diagnoses

Figure 1 Haemangioma of the right inferior turbinate (preoperatively).

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included also rhinosporidiosis and cavernous haemangioma. Rhinosporidiosis presents as a 'strawberry-like' mass, a friable vascular lesion with numerous tiny white epithelial spores on the surface. Cavernous haemangioma, is even rarer in the nose, and consists of closely packed dilated blood-filled vascular channels lined with flattened endothelial cells.⁶

TREATMENT

In theatre she underwent endoscopic resection of the tumour and inferior turbinate as well as endoscopic medial maxillectomy (figure 2). Histological analysis showed an ulcerated and infarcted polypoidal lesion, with a proliferation of small to medium sized vessels and an inflamed fibrous stroma. There was no evidence of atypia, and a diagnosis of benign haemangioma was performed. Postoperatively she was prescribed coamoxiclav and Stérimar nasal spray to reduce the chance of her developing a postoperative sinus infection, and to prevent nasal crusting respectively.

OUTCOME AND FOLLOW-UP

She was followed up 3 weeks later in clinic and was found to have made a good recovery, albeit with some crusting of the right nasal cavity, which was removed under local anaesthesia. A further follow-up 3 and 9 months later and a repeat CT scan showed no recurrence of this benign tumour.

DISCUSSION

Sinonasal LCH is a rare condition and as such there are relatively few reported instances of it occurring. The first case of LCH in the literature was in 1897, when Poncet and Dor named it human botryomycosis. They described vascular masses which appeared on the fingers and arms of four patients, resembling the strawberry-like lesions which formed on the scrotum of a castrated horse, and were believed to be caused by the fungus botryomycetes.⁷ There were called pyogenic granuloma by Hartzell⁸ in 1904, as he thought they were a non-specific granulation tissue reaction to a pyogenic infection. It was not until later in the 1980s when Mills, seeing little in the way of evidence for an infective cause, described them as LCH due to their typical lobular capillary arrangement at the base.²

Which of these terms represents the correct nomenclature is difficult to say and the topic of some debate. If it is a true neoplasm,

then the term, haemangioma should be used; however, if it is a reactive tumefaction, then pyogenic granuloma would be the more appropriate name. As there is a lack of concordance between cases to implicate a common causative agent, or local trauma, as the cause of the lesions, it is more likely to be a true neoplasm, albeit it may be that there is a causative agent responsible for them, which has yet to be elucidated. This question is somewhat of a moot point however, as its method of treatment and prognosis is widely agreed on, in that it is a benign lesion for which surgical resection is the cure.⁹ Furthermore whether the haemangioma is of capillary, cavernous or mixed type is of no clinical significance, it relates only on to the dominant vessel size seen on microscopy of the lesion.^{6 10}

Smith *et al*¹¹ found 38 instances of this condition in their institution from 1989 to 2009. The presenting symptoms commonly included epistaxis (76% of patients) but otherwise showed little concordance: 36% experienced nasal obstruction and only 3% pain. With haemangioma of the maxillary sinus, patients have been noted to present with proptosis and facial numbness.¹²

Epidemiology

Smith *et al*¹¹ found an equal prevalence of LCH between men and women; however, 29.4% of the women were currently, or had recently been, pregnant. There was also an association with previous trauma to the area where the haemangioma developed in 11.8% of patients. Although this data provides some insight into the presentation and epidemiology of sinonasal haemangioma, it is of somewhat limited value as it included few patients and it was a retrospective data analysis.

LCH has been found to occur in all age groups, although it appears more frequently in patients in their 30s. In the adult population LCH is more prevalent in women, possibly due to the increased risk of developing LCH during pregnancy, while in children it is more common in men.¹³

Diagnosis/imaging

The use of CT scans has been purported as a method of diagnosing LCH, without requiring a biopsy to be taken, however, when the lesion is large it can be difficult to distinguish it from other neoplastic and inflammatory conditions. This is further illustrated in the case reported, where the opinion of the radiologist on the CT scan was that of an aggressive localised tumour consistent with an inverted papilloma or possibly a squamous cell carcinoma. This led to the patient having an urgent resection biopsy for suspected malignancy, which meant that the haemangioma was quickly removed and the correct diagnosis performed.

Pathophysiology

The exact aetiology of LCH is not known, however, correlations are seen with the use of oral contraceptives, trauma, post-operative nasal packing, microscopic arteriovenous malformations, cytogenetic abnormalities, viral oncogenes, the production of angiogenic growth factors⁴ and pregnancy; LCH has been described as a pregnancy tumour, due to its increased prevalence in pregnancy.¹⁴ The haemangiomas which form in pregnant women regress after parturition, indicating a role for the hormones of pregnancy in their proliferation.³ Local trauma has been noted to have a positive association with the formation of LCH; however, the incidence of this is small, varying from 4.5% to 15%, although an increased incidence of LCH has been seen in those patients who frequently pick their noses.^{15 16} It is also of note that the lesions characteristically appear on the

Figure 2 Inferior antrostomy after complete endoscopic excision of the right inferior turbinate.

Patient's perspective

Date	Comment
Late July 2012	I first presented with an obstructed right nostril and yellow secretions coming into the back of my throat <i>Possible cause: In early July 1 received a striking blow to the right side of my nose and cheek from an autistic pupil at the school where I worked as a nurse</i>
August 2012	I was now presenting with continuous severe headache/migraine, obstructed right nostril, right eye watering, right side of nose tender, upper jaw ache, clear nasal secretions but blood stained yellow secretions coming into the back of my throat
5th September 2012	I was seen by a locum general practitioner (GP) who diagnosed sinusitis (my right nostril was not examined) and prescribed amoxicillin and Beconase spray which was too excruciatingly painful to use
Late September 2012	I went on holiday with my daughter who is a GP and witnessing my severe headaches and nasal/sinus distress advised me to see my GP and ask for an immediate referral and scan
12th October 2012	I saw my own GP who could see a tumour when examining my right nostril
17th October 2012	I was seen by the Ear, Nose, and Throat (ENT) Department at Derriford Hospital. Using an endoscope a query inverted papilloma was seen. The doctor ordered MRI and CT scans and prescribed Augmentin
31st October 2012	The scan revealed a medium sized mass in my nasal cavity which could possibly be malignant
8th November 2012	The operation to remove the mass went well. Postoperatively I had an absorbable nasal pack in situ and was prescribed Augmentin, analgesics and Sterimar spray
26th November 2012	My ENT consultant, telephoned and informed me that the laboratory test results showed that he had removed a benign haemangioma
Postoperative care	I have had regular appointments with my ENT consultant since the operation. He has examined my nasal cavity with an endoscope at each visit and prescribed further courses of Augmentin, doxycycline and nasonex spray when necessary
5th July 2013	A further CT scan revealed no significant abnormality

Additional comments:

Postoperatively, with the aid of Sterimar spray, very large crusts were 'blown' with difficulty from my right nostril. The ENT consultant also surgically removed some crusting under local anaesthetic. After 17 months, I am still producing green crusts albeit much smaller and now only every 4–6 weeks. I continue to use Sterimar spray daily.

I am still suffering discomfort from thick green/yellow/clear secretions (the colour varies with the timing of the shedding of the crust) which come from my sinuses and collect in the back of my nose/throat. I have to constantly work to clear these secretions.

At night the secretions seem to be more troublesome and on waking I have to cough persistently to clear my upper respiratory passages.

Although I can now breathe very easily through my right nostril, I find it difficult to blow secretions from it—the secretions pass directly down the back of my throat.

Hopefully I am still in the healing process and the above problems will dissipate with time.

I shall be eternally grateful to my ENT Consultant and his Registrar for their excellent medical/surgical expertise in the handling of my condition. My GP referral took only 4 days! At every appointment I have been treated with great care, attention and professionalism. At the Freedom Unit, on the day of my operation my ENT Consultant and his Registrar took time to fully explain the procedure. The anaesthetic consultant instilled confidence and the theatre staff were very reassuring. I felt I could not be in safer hands. The recovery room staff was diligent and very concerned for my level of comfort.

It was extremely thoughtful of the ENT Consultant to inform me immediately he knew that the excised tumour was benign.

I commend the Otorhinolaryngology-Head and Neck Surgery (ORL-HNS) Department for its wonderful work.

Many, many thanks.

Kind regards,

The patient

anterior part of the inferior turbinate,⁶ or in Kiesselbach's plexus, areas of the nose which are more easily accessible and therefore more prone to trauma.³

Although local trauma and nose-picking have both been associated with LCH formation, they are only reported as the probable cause in a small number of cases. Besides which, local trauma and nose-picking are quite common phenomena in the general population, and so if they were a significant cause of LCH, then the incidence of the lesion would be expected to be far higher than that which is seen.

Sinonasal LCH is seldom found as an incidental finding, as patients tend to present to their doctor swiftly after the onset of

symptoms: nasal obstruction and epistaxis.⁹ This is beneficial as the disease will be caught at an early stage. However, this is not always the case as we saw in our report. It can still grow to a considerable size with local tissue destruction. Malignant lesions can present in a similar manner, and a conscientious physician will seek to exclude these rather than misdiagnose them as LCH.

This case report will raise awareness of LCH as a potential diagnosis in patients presenting with unilateral nasal obstruction and epistaxis. Although it may not be a common condition, or a particularly hazardous one if misdiagnosed, it is a more palatable diagnosis if it can be confirmed by biopsy or nasal

endoscopy, for patients who would otherwise fear that they had a malignant lesion. However, whenever the patient presents with rapidly enlarging nasal mass or bony erosion, then radiological tests must be undertaken to rule out malignancy.¹³

The rarity with which LCH presents in the sinonasal region can lead to uncertainties in the diagnosis. A delayed diagnosis can lead to rapid haemodynamic imbalance if the mass ruptures and epistaxis occurs, due to its highly vascular and friable nature.⁹ The diagnosis of LCH should therefore be considered from the outset in cases of unilateral nasal obstruction and epistaxis. Further education about this lesion is deemed important as preoperative embolisation may be needed for a safe operation.

Learning points

- ▶ Lobular capillary haemangiomas are benign vascular tumours which are common in the head and neck.
- ▶ They are rare in the paranasal sinuses where they present with epistaxis and unilateral nasal obstruction.
- ▶ They can be hard to diagnose and are frequently mistaken for malignant lesions.
- ▶ They can be cured surgically via endoscopic resection.
- ▶ Further education about this lesion is deemed important as preoperative embolisation may be needed for a safe operation.

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