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Clinical Record

Granular cell tumour arising from the Kiesselbach's area of the nasal septum

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Abstract

An extremely rare case of a granular cell tumour arising from the right Kiesselbach's area (Little's area) of the nasal septum is reported. A 69-year-old Japanese woman consulted our clinic and her chief complaints were of continuous serous discharge, stuffiness and occasional slight bleeding from the right nasal cavity. Fibrescopy showed a multilocular mass, which was provisionally considered a nasal polyp. Surgical excision was attempted. During surgery, the tumour shrank markedly following local application of adrenaline, suggestive of hypervascularity. The tumour was successfully excised by careful dissection after cauterisation of the mucosa surrounding the tumour. Histopathological examination revealed morphological features of granular cell tumour immunopositive for S-100 protein. This is the first report of granular cell tumour arising from the Kiesselbach's area in the English literature. The eccentric behaviour of the tumour and the management of a granular cell tumour arising from this area are discussed, together with a literature review.

Key words: Granular Cell Tumour; Nasal Septum; Epistaxis

Introduction

A granular cell tumour is an uncommon benign tumour that can be found in almost all body regions but particularly in the skin and oral cavity, especially the tongue.[1] However, it is rarely found in the nasal cavity and based on a 1960–2005 Medline database search, only one case has been reported.[2] Here we present a rare case of a granular cell tumour arising from the Kiesselbach's area (or Little's area) of the nasal septum. The patient suffered from frequent episodes of nasal bleeding and the tumour shrank in size after topical application of adrenaline. These clinical features are commonly associated with haemangioma, and have not been observed in the previously reported cases of granular cell tumours.[1–4,7,8,12] That it originated from the most common site of epistaxis may explain the tumour's unique clinical behaviour.

Case report

A 69-year-old Japanese woman presented to our clinic on 17 October 2005 complaining of continuous serous discharge, stuffiness and occasional bleeding from the right nasal cavity for several years. She had already visited another otolaryngologist and was told that the cause of those symptoms was a polyp occupying the right nasal cavity. Oral administration of antihistamine had already been attempted and resulted in no improvement. She also reported slight nasal bleeding after suction of the right nasal cavity in the former clinic. Nasal fibrescopy performed at our clinic showed a multilocular tumour comprising of at least two spherical masses with a smooth surface obstructing the nasal cavity (Figure 1). The tumour was pale pink in colour and almost similar to that of the nasal mucosa. The overall impression was that the lesion was not a nasal polyp. Inspection of the left nasal cavity, pharynx, larynx and oral cavity was unremarkable. Palpation of the neck disclosed no lymphadenopathy. The patient consumed no alcoholic beverage and had no history of smoking.

Computed tomography (CT) showed an oval mass (19 mm × 14 mm) in the right nasal cavity (Figure 2). The base site of the tumour was still unclear from these findings. To confirm the diagnosis and to resect the tumour an operation was performed on 24 November 2005 under local anaesthesia. Slight bleeding occurred when the tumour surface was touched with the tip of the forceps, but it stopped spontaneously. Our initial plan was to identify the stalk of the tumour by application of electric cautery, followed by resection of the tumour. Following our usual procedure for nasal surgery we inserted a few pieces of cotton impregnated with 0.02 per cent adrenaline and 4 per cent lidocaine in the right nasal cavity for approximately 10 minutes. This resulted in marked shrinkage of the tumour, probably due to the effect of the adrenaline, and the tumour mass changed to a small notch on the nasal septum mucosa (Figure 3). This site corresponded to Kiesselbach's area, the most common site of epistaxis. Based on this behaviour and the history of occasional bleeding, a hypervascular tumour such as a haemangioma was suspected. However, the colour was different from a haemangioma, which normally presents as a red or purple coloured tumour. Based on the new finding, the operation strategy was altered and attention was focused on preventing uncontrollable bleeding. Before resection

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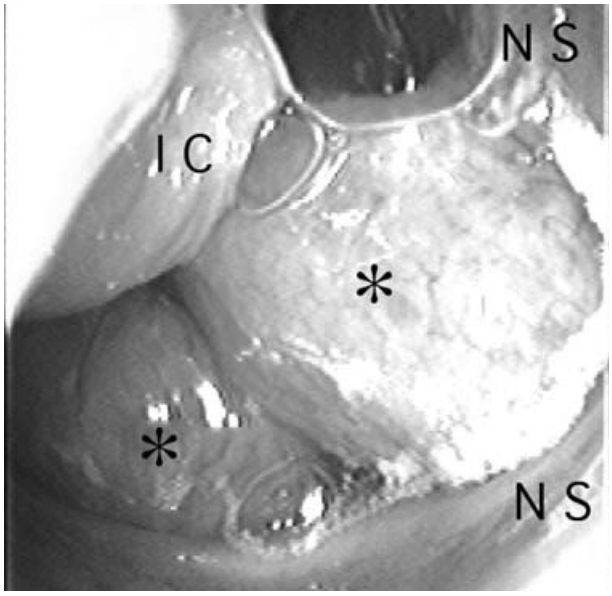

FIG. 1

Fibrescopy of the right nose on initial consultation. Note the spherical masses with smooth surface (asterisk), which obstructed the nasal cavity. IC = inferior nasal concha; NS = nasal septum

of the tumour, the mucosa around the tumour was cauterised using an electric bipolar coagulator. The cauterised mucosa was dissected carefully by scalpel and scissors. The base of the tumour, which was somewhat firm and attached to the cartilage of the nasal septum, was exfoliated with a blunt curette. Bleeding was well controlled during the operation. Although the margin of the resected specimen was cauterised, the majority of the main part of the tumour remained intact and looked fully suitable for histopathological examination. A gelatine sponge sheet was applied over the site of excision. At the last follow up examination three months after the operation, no tumour recurrence was noted. The patient no longer suffers from nasal discharge, stuffy nose or epistaxis and is followed up in our clinic.

Histopathological examination showed tumour cells arranged in nests with eosinophilic cytoplasm containing pale granules (Figure 4). These granules were positive on periodic acid-Schiff stain and Alcian blue stain. Interstitial vessels were somewhat abundant. The nuclei were dense

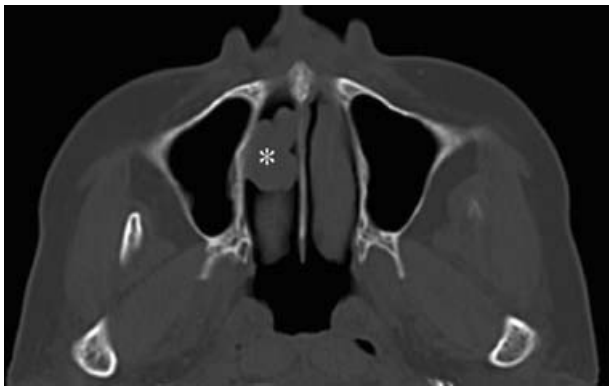

FIG. 2

CT scan image showing ovoid-shaped tumour (asterisk) at the level of the nasal cavity.

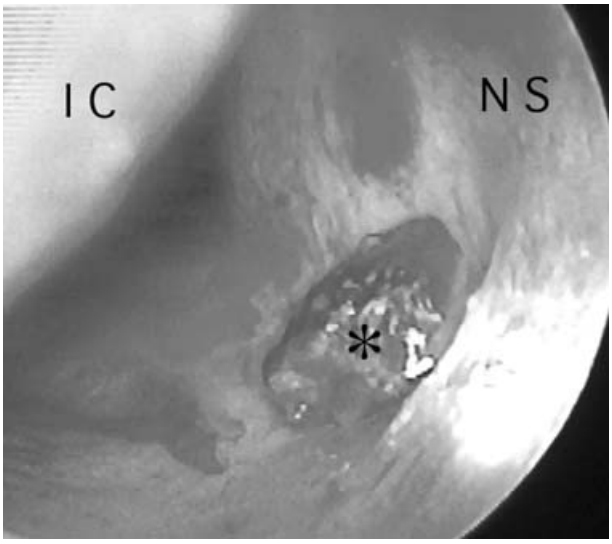

FIG. 3

Marked shrinking of the tumour following application of adrenaline to the nasal septum (asterisk). IC = inferior nasal concha; NS = nasal septum

and showed no atypia. No mitosis was seen. Immunohistochemical staining with S-100 protein, a marker for Schwann cells, was positive (Figure 5). Cytokeratin markers CAM 5.2 and AE1/AE3 were negative. Vascular markers CD34 and CD31 were also negative. The above findings were consistent with a granular cell tumour.

Discussion

A granular cell tumour is an uncommon lesion with a site predilection for the skin and oral cavities, particularly the tongue, though it can be found in all organs and tissues.[3,4] The report of five cases of granular cell tumour by Abrikossoff in 1926, which included three cases of the tongue, one case of lip and one case of the thigh, is often referred to as the first description.[5] However, earlier in 1854, Weber reported a case arising from the tongue.[6] The granules in the cytoplasm observed by haematoxylin and eosin staining is characteristic and supportive of the diagnosis.[3] The origin of the tumour was formerly suspected as striated muscle but it is currently considered to be of nerve origin, though this is still a controversial issue.[4] S-100 protein positivity is characteristic for the diagnosis of granular cell tumour in all cases except those of congenital gingival tumours.

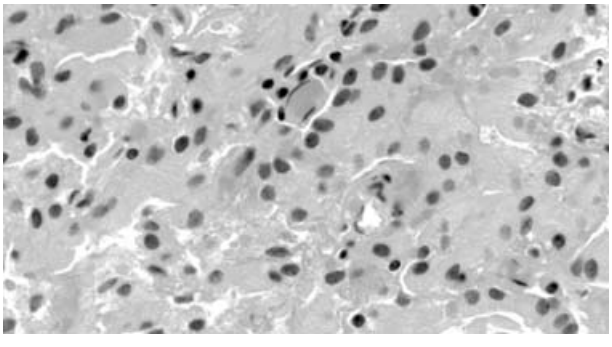

FIG. 4

High-power view of tumour cells (H&E; ×400).

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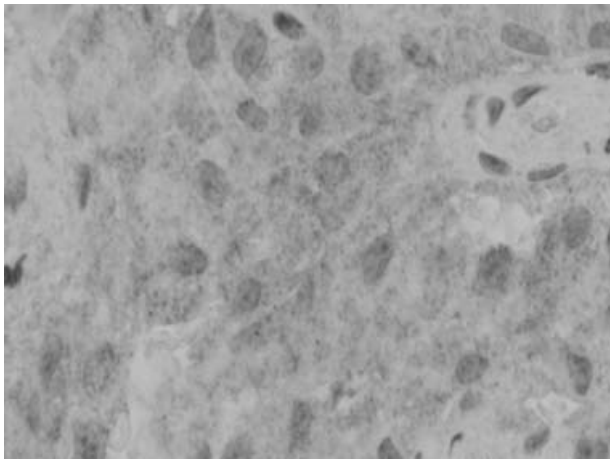

FIG. 5
Photomicrograph of tumour cells immunoreactive to S-100 (H&E; ×400).

The incidence of malignant changes varies among reports from 1–7 per cent.[4,7,8] The tumour is more likely to be found in females and is more prevalent in those aged 30–50 years.[1,4]

Granular cell tumours are rarely found in the nasal cavity and to our knowledge; only one case had been reported previously in the English literature.[2] In that case, the tumour was found in the left nostril and extended to the anterior part of the nasal septum, a site somewhat different from our case. Thus, our case is the first report of a granular cell tumour arising in the middle of the Kiesselbach's area.

A granular cell tumour is not generally exophytic, rather frequently endophytic and usually exhibits permeative growth. In our case, the tumour was spherical and initially misdiagnosed as a polyp in another clinic. Because of the paucity of granular cell tumours arising in the nasal septum, it is difficult to resolve the cause of this uncommon exophytic appearance. One speculation is that this is a reflection of the unusual site of this tumour. This is analogous to the fact that papillomas arising in the nasal septum are frequently fungiform, whereas they are more often endophytic when arising elsewhere in the sinonasal region.[9,10] Deep infiltrative permeation of the tumour might be halted by some unknown factors in the nasal septum.

In general, a granular cell tumour is not considered haemorrhagic. Our case was clinically interesting in that the tumour presented with a bleeding tendency and showed marked shrinkage following adrenaline appliance. This behaviour is often seen in hypervascular tumours such as a capillary type haemangioma, also known as a 'bleeding polyp' of the nasal septum.[11] However, the appearance of the tumour in our case was completely different from that of haemangioma, which normally appears red or purple in colour.[11] Why did the tumour behave like a haemangioma? Again, we presumed that this is a consequence of the unusual location of the tumour in our case. The Kiesselbach's area is the most common site of nasal bleeding due to the rich vascular plexus and tumour development in this area leads to this feature. In fact, histopathological examination of the resected specimen of our case showed relatively abundant vessels, which is an uncommon finding in the previously reported granular cell tumour cases.[1–4,7,8,12] This pathological finding seems to support our speculation. We assume that because the tumour originated in this vessel rich site, the tumour tissue intermingled with the thick vessel network of the Kiesselbach's area as the tumour proliferated, with subsequent growth as a haemorrhagic tumour with high vascularity.

In general, surgical excision is the first choice of treatment for a granular cell tumour arising from common sites such as the skin. The extent of surgery ranges from a simple local enucleation to excision with a wide margin. In some cases, extirpation with a CO_2 laser is reported to be successful.[1,8] Recurrence of the tumour is reported in 8–12 per cent of the reported cases, which is attributed to an insufficient excision.[3] Due to the rarity of granular cell tumours arising in the nasal cavity, there are no accepted standards with regard to the management. In our case, we cauterised the mucosa surrounding the tumour and excised the tumour mass. However, one case of granular cell tumour arising from the maxilla was reported to exhibit expansive growth together with destruction of the surrounding bone.[12] Careful monitoring of the excision site is needed in our case for the detection of recurrence and possible destructive tendency.

- **A previously unreported case of a granular cell tumour arising from the right Kiesselbach's area (Little's area) of the nasal septum is presented**
- **The tumour was also unique based on a bleeding tendency, shrinkage following adrenaline application and hypervascularity on histopathological examination, which are uncommon findings compared with previously reported granular cell tumours**
- **The cause of hypervascularity and the management of this tumour are discussed**

In summary, we reported a very rare case of a granular cell tumour arising from the Kiesselbach's area of the nasal septum, presenting with a bleeding tendency. The tumour was successfully excised after cauterising the surrounding mucosa. Granular cell tumours should be considered in the differential diagnosis of similar cases encountered in the future.

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