Nasal leiomyoma presenting as sphenopalatine neuralgia: a case report and literature review

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Abstract

Leiomyoma of the nasal cavity constitutes less than 1% of all leiomyoma cases and is known to occur predominantly among the adult female population.[1] Traditionally, intranasal leiomyoma presents with nasal obstruction and epistaxis. Herein, we report a case of solid-pattern leiomyoma arising from the right inferior turbinate, presenting exclusively as severe right jaw and neck pain, made worse by neck movements, swallowing, and even sneezing. Pain was associated with facial spasm and had significant impact on her quality of life. Endoscopic excision of the tumour successfully alleviated the patient's symptoms, requiring no further management. An extensive literature search revealed that intranasal leiomyoma could be successfully managed by endoscopic excision with no recurrence. We highlight the importance of a thorough intranasal endoscopic examination in all patients with persistent neuropathic pain of the head and neck.

Introduction

Leiomyoma is a benign smooth muscle tumour most commonly occurring in the uterus and gastrointestinal tract. Intranasal leiomyoma is rare and constitutes less than 1% of all reported cases.1 Intranasal leiomyoma typically has a similar presentation to other benign tumours arising from the nasal cavity. Symptoms include nasal obstruction, nasal discharge, unprovoked epistaxis, and facial pain.[2] We report a case of intranasal solid pattern leiomyoma presenting solely as sphenopalatine neuralgia in which the patient was treated for chronic neuropathic pain for over a five-year period. Computed tomographic scanning of the head and neck region for possible Eagle's syndrome (characterized by facial and/or neck pain radiating to the ear and throat, caused by irritation of neurovascular structures by an elongated styloid process) incidentally detected a solid lesion within the right nasal cavity, with biopsy confirming the diagnosis of leiomyoma.[3] Surgical excision of intranasal leiomyoma effectively relieved her symptoms, and the patient is well thereafter. In this case report, we highlight the importance of conducting more vigorous investigations for neuropathic pain in the head and neck region to rule out the presence of intranasal leiomyoma as treatment is straightforward and with excellent outcome.

Case Report

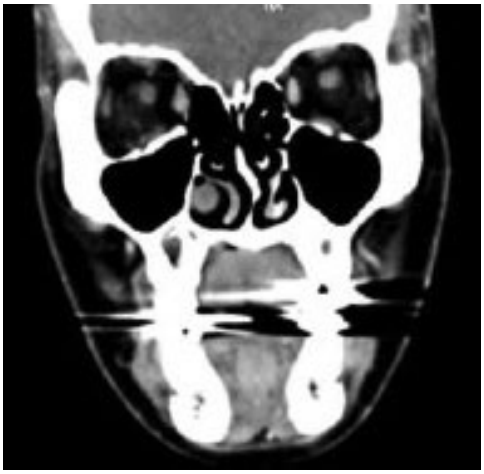

Figure 1. Coronal computed tomography image showing opacity over the right inferior turbinate

A young female with underlying antiphospholipid syndrome presented with a five-year history of excruciating right-sided facial pain, typically over the angle of the jaw, associated with tearing and occasionally facial spasms. According to the patient, the right-sided jaw pain was provoked by head and neck movements, swallowing, and sneezing. The pain was debilitating, with a significant negative impact on her marriage, career advancement, and overall quality of life. The patient had received multiple medical consultations and was treated for trigeminal autonomic cephalgia with various

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analgesics, including NSAIDs, antiepileptics (Carbamazepine, Pregabalin, Lamotrigine), an antidepressant (Fluoxetine), steroids, chiropractic treatment and traditional medications. None of the treatments relieved her symptoms. Additionally, the patient denied any recurrent nasal, throat or ear symptoms. No other neurological or constitutional symptoms were noted. During her visit to an oral maxillofacial surgeon, an orthopantomogram, a panoramic radiograph of the teeth, maxilla and mandible, revealed an asymmetric styloid process. Hence, a computed tomography (CT) of the neck was performed to exclude Eagle's syndrome. The CT revealed bilateral styloid processes to be of normal length (less than 3cm), with the right side measuring 1.2 cm. There was, however, an incidental finding of a well-defined nasal opacity inferior to the right inferior turbinate (Figure 1).

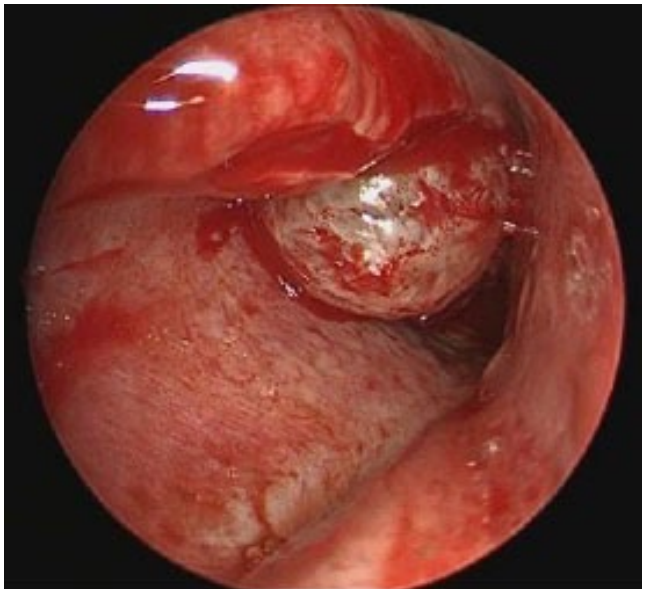

Figure 2. Endoscopic view of the soft tissue mass in the right nasal cavity seen after partial inferior turbinectomy

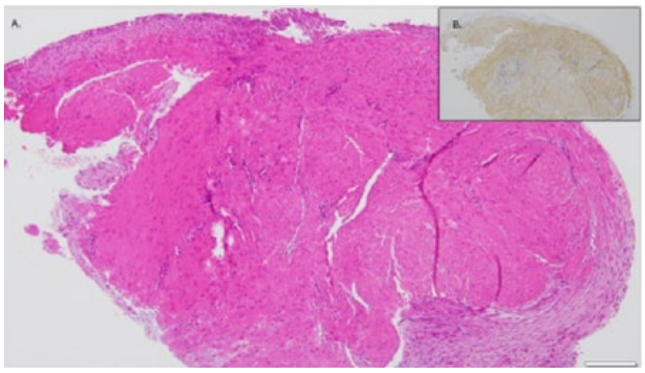

Figure 3. Endoscopic view of the soft tissue mass in the right nasal cavity seen after partial inferior turbinectomy

Endoscopic examination confirmed the presence of a vascular, polypoidal mass arising from the anterior aspect of the right inferior turbinate. Biopsy sent for histopathological examination was consistent with leiomyoma. Eventually, the patient underwent partial turbinectomy and excision of the right intranasal tumour via an endoscopic approach (Figure 2). Intra-operatively, a soft tissue mass about 1x1 cm was seen arising from the right lateral nasal wall, inferior to the inferior turbinate. Histopathological evaluation of nasal specimen confirmed it as a leiomyoma of solid pattern subtype (Figure 3).

No intra-operative or post-operative complications were noted. Interestingly, following the excision, the pain in the jaw subsided. Subsequent follow-up revealed no recurrence of symptoms, and the patient remains symptom-free to date, which is just over two years post-operatively.

Discussion

The sphenopalatine (or pterygopalatine) ganglion is located in the pterygopalatine fossa, and lateral to the sphenopalatine foramen. It gives out branches which have secretomotor and sensory functions to the mucous membrane of the nasal cavity, oral cavity, soft palate and parts of the pharynx. In 1913, Dr Greenfield Sluder first described sphenopalatine ganglion neuralgia, which he described as facial pain around the nose and eye, in the upper jaw and teeth, extending backwards under the zygoma to the ear and mastoid, to the occiput, neck, shoulder, which may extend along the arm down to the fingers.[4] Sphenopalatine neuralgia (also known as Sluder's neuralgia) is caused by irritation of the sphenopalatine ganglion from direct compression (for example, by an intranasal mass or a deviated nasal septum), infection, or involvement of nerve endings during fibrosis of nasal mucosa.[5]

Intranasal leiomyoma is a benign, slow-growing tumour arising from the vascular smooth muscle cells of the nasal cavity. The first intranasal leiomyoma case was reported by Maesaka et al in 1966.[6] Head and neck leiomyoma constitutes less than 1% of all leiomyoma. Among them, only about 2% are found within the nasal cavity, most of which arise from the anterior part of the inferior turbinate. Similarly, in our patient, the mass was located in the same region.

A literature search demonstrated that intranasal leiomyoma is more frequently seen in the adult female population.[7-13] The most common clinical presentations of intranasal leiomyomas include nasal obstruction, nasal discharge, unprovoked epistaxis, and, occasionally, facial pain. It can be easily detected during endoscopic examination of the nose as a pink-reddish polypoidal mass occupying part or whole of the nasal cavity. It may arise anywhere in the nasal cavity, including the lateral nasal wall (middle turbinate or inferior turbinate), nasal septum, nasal floor, as well as the nasal vestibule.[2,7-14] To the best of our knowledge, this is the first case report of intranasal leiomyoma presenting with symptoms mimicking sphenopalatine neuralgia without any associated nasal symptoms. We posit that the symptoms were most likely caused by irritation of the sphenopalatine ganglion or its branches.

The most commonly employed radiological investigation to evaluate intranasal mass is computed tomography of the nose and paranasal sinuses. On CT images, intranasal leiomyoma presents as a contrast-enhanced space-occupying homogenous mass without evidence of bony destruction.[1,9,11] A review of 10 case reports between 1981-2015 showed that intranasal leiomyomas were excised surgically.[1-2,7-14] Endoscopic trans-nasal excision was advocated for most patients, with lateral rhinotomy performed on patients with exceptionally large masses. Intraoperatively, intranasal leiomyoma may arise from different parts of the nasal cavity, including the lateral nasal wall, either of the three turbinates, as well as the nasal septum.

The most common histopathological subtype found in the

head and neck region is solid pattern leiomyoma (71%), followed by angioleiomyoma (27%) and epithelioid leiomyoma (1.2%).[15] Solid-pattern leiomyoma was noted in our patient. Sinonasal angioleiomyoma has been reported to have a much higher prevalence among the female population compared to the male population (3.75:1 female: male ratio). This may be attributed to the fact that vascular Leiomyoma is hormonal dependent, as shown by the detection of progesterone receptors in published literature. Diagnosis of a leiomyoma could be confirmed by the detection of smooth muscle actin, desmin and vimentin during immunohistochemical staining.[16]

Complete recovery was reported for all patients without recurrence after surgical excision. In our case, an immediate and permanent resolution to sphenopalatine neuralgia was achieved after the mass was excised.

Conclusion

Clinical practitioners, especially primary care doctors, should familiarize themselves with the possible causes of non-resolving facial and neck pain, one of which could be intranasal Leiomyoma which is easily treatable. Referral to an otorhinolaryngologist for endoscopic examination of the nose to rule out compression or irritation of sphenopalatine ganglion or its branches is justified.

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