Isolated oncocytic sinonasal papilloma of the sphenoidal sinus

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SUMMARY

The ectodermally derived mucosa of the nose and paranasal sinuses may give rise to three morphologically different papillomas referred to individually as exophytic papilloma, inverted papilloma (IP) and oncocytic sinonasal papilloma (OSP). OSP is the rarest of the three different types, frequently affects the maxillary sinus, ethmoid sinus and nasal cavity while sphenoidal sinus involvement is exceedingly rare. The tumour has no gender predilection and has a wide age range with the majority above 50 years of age. Clinically, its behaviour parallels IP due to local recurrence and tendency to malignant transformation. Microscopically, the tumour shows multilayered eosinophilic epithelium with exophytic and inverted growth patterns. We report a case of OSP of the sphenoidal sinus, which, to the author's knowledge, is one of the few cases of isolated sphenoidal sinus OSP reported in the English literature. In this article, we describe how the patient presented, was diagnosed and how it was managed.

BACKGROUND

Oncocytic sinonasal papilloma (OSP), also known as cylindrical and columnar cell papilloma (CCP), is a rare type of sinonasal papilloma and represents 3%–5% of all sinonasal papillomas.[1–3] Barnes and Bedetti in 1984 demonstrated that the epithelial cells of the cylindrical cell papilloma are the oncocytes which arise from the sinonasal respiratory epithelium hence the term OSP.[1] The ectodermally derived mucosa of the nose and paranasal sinuses may give rise to three morphologically different papillomas referred to individually as the exophytic papilloma (EP), inverted papilloma (IP) and OSP or collectively as Schneiderian papillomas.[2 4 5] The reported incidence of papillomas of the nasal cavity and paranasal sinuses varies from 0.4% to 4.7% of all nasal and paranasal sinus tumours[2 6] and nasal polyps for instance, are stated to be 25–60 times more common than papillomas.[7] Hyams reported in a study of 315 cases in 1971 the proportion of three types of papillomas as EP 49.5% and IP 47.3%, and CCP 3.2%[2] and in a Danish series of 82 patients in 1995 reported 23%, 70% and 6%, respectively.[5] OSP being the rarest of three types of papillomas occurs mostly in patients over 50 years of age and are equally distributed between sexes.[3 4 8 9] They occur almost always unilaterally, usually in the maxillary sinus, nasal cavity and ethmoidal sinus, whereas its exclusive location within the frontal or sphenoidal sinus is exceedingly rare (only 5% of all paranasal sinuses).[1 2 10 11] The morbidities of the three types of papillomas are quite different and it is important to note that between 4%–17% of the total OSP have been associated with malignant transformation[2 4 8 9] with invasive squamous cell carcinoma being the most frequently reported tumour.[3 4 6] Clinically, behaviour of the OSP is comparable to IP for local recurrence (33%–40%)[3 12] and smokers having more recurrence rate than non-smokers (75% vs 31%) as well as the tumours located in the sinuses having more recurrence rate than those located in the nasal cavity (45% vs 29%).[11] The clinical features of sinonasal OSPs are nonspecific and most of the patients present with unilateral nasal obstruction and a soft-tissue mass while rhinorrhoea, intermittent epistaxis, facial pain and headache may also occur in some patients.[4 8 10 13] These symptoms often persist for several months to a few years because of the slow growth of OSP.[2 4 14] Advanced imaging such as CT and MRI has enhanced the ability of the surgeon to diagnose, localise and resect the tumour with greater precision than ever before. Surgical treatment (endoscopic endonasal or external approach) depends on the size and mainly localisation of the tumour. In this article, we report a case of an isolated sphenoidal sinus OSP with positive FDG (fluorodeoxyglucose) uptake in positron emission tomography (PET)/CT, which is a rare finding as mentioned above. A few case reports regarding the findings of OSP at PET/CT have been documented in the English literature.[13 15–17] To the best of our knowledge, this is one of the few cases reported in the literature with isolated sphenoidal sinus involvement[13] and the first case reported in the English literature with bilateral sphenoidal sinus involvement. According to the previous literature review, there is another case report of an OSP originating from the sphenoidal sinus and extending to the nasal cavity on the unilateral side, though the

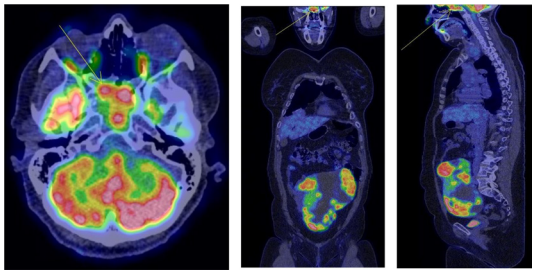

Figure 1 Positron emission tomography-CT scan (axial, coronal and sagittal) showing positive fluorodeoxyglucose uptake in the sphenoidal sinus.

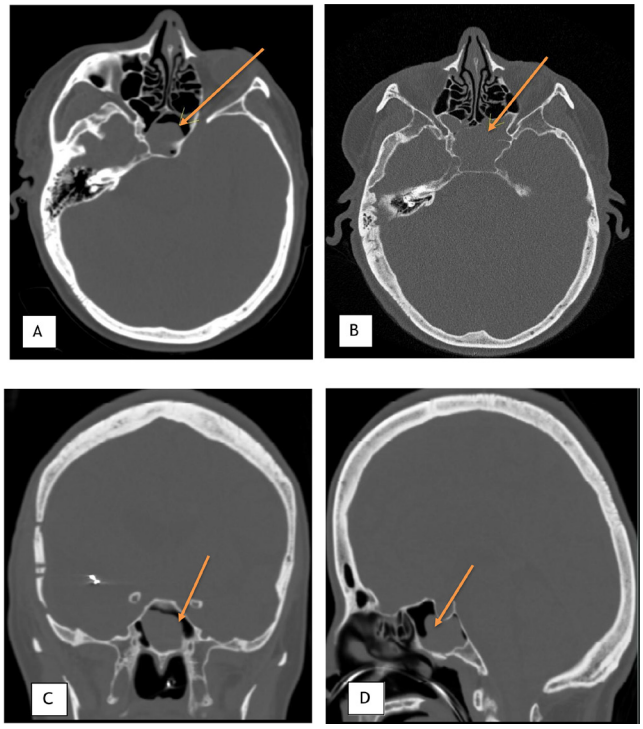

Figure 2 (A) shows the polypoid lesion in the left sphenoidal sinus in 2014 in axial view (arrow), (B) shows the lesion filling the bilateral sphenoidal sinus in 2023 (arrow) and (C and D) show the polypoid lesion in the left sphenoidal sinus in 2014 in coronal and sagittal view.

epicentre of the tumour can be seen in the nasal cavity according to the figures of the article.[18]

CASE PRESENTATION

Here we report a rare case of OSP of the sphenoidal sinus which was diagnosed in 2023. The tumour was first highlighted as a positive FDG-uptake polypoid lesion with possible bony destruction in the sphenoidal sinus after a PET-CT scan (figure 1) which was done with a suspicion of disseminated ovarian carcinoma in a female patient who was in her mid-50s.

The patient did not complain about any of the related symptoms of sinonasal papillomas, such as headache, visual disturbance or nasal symptoms and was referred to the ear, nose and throat department for further management. Retrograde evaluation of the previous CT scan of the cerebrum, which was done after a subarachnoid haemorrhage due to a ruptured left middle cerebral artery aneurysm in 2014, confirmed a relatively smaller polypoid lesion in the left sphenoidal sinus (figure 2A). No previous surgical interventions of the nasal cavity or paranasal sinuses have been reported and no further interventions were done regarding the polypoid lesion in the sphenoidal sinus.

Supplementary MRI was done after 15 days of PET-CT and the bilateral sphenoidal sinuses appeared to be filled with inhomogeneous contrast-enhancing soft tissue tumour with thinned and possibly missing sinus wall. However, without clear signs of penetration into the surrounding soft tissue compartments and no clear bone invasion (figure 3A–D). The tumour has a characteristic grapelike structure on MRI and is isointense on T1, mildly hyperintense on T2-weighted sequence and heterogeneously and moderately enhancing after i.v. contrast.

CT paranasal sinus was done on the same day after MRI, considering suspicion of an isolated sphenoidal sinus inverted papilloma, which showed the soft tissue tumour with possible bone destruction (figure 2B).

It was decided on a wide endoscopic resection of the lesion and computer-assisted surgery - functional endoscopic sinus surgery with a block tumour excision, with successful removal of the complete tumour after a few days. Results of the intraoperative frozen section were indicative of sinonasal papilloma with exophytic and endophytic (inverted) growth. The postoperative histological diagnosis was consistent with OSP.

Histopathology revealed a sprawling epithelium that was partially coated with respiratory epithelium and cytoplasm. It was partially eosinophilic and granular (oncocytic), which grew partially endophytic into the stroma, forming an epithelial islet and partially exophytic, forming papillary structures with fibrovascular stalks. Lymphocyte cells, PMN leukocytes and mucin-filled cavities in epithelium were noted. Partially densely encapsulated groups of lymphocyte cells, plasma cells, histiocytes, PMN leucocytes and seromucinous glands were seen beneath the connective tissue stroma (figure 4).

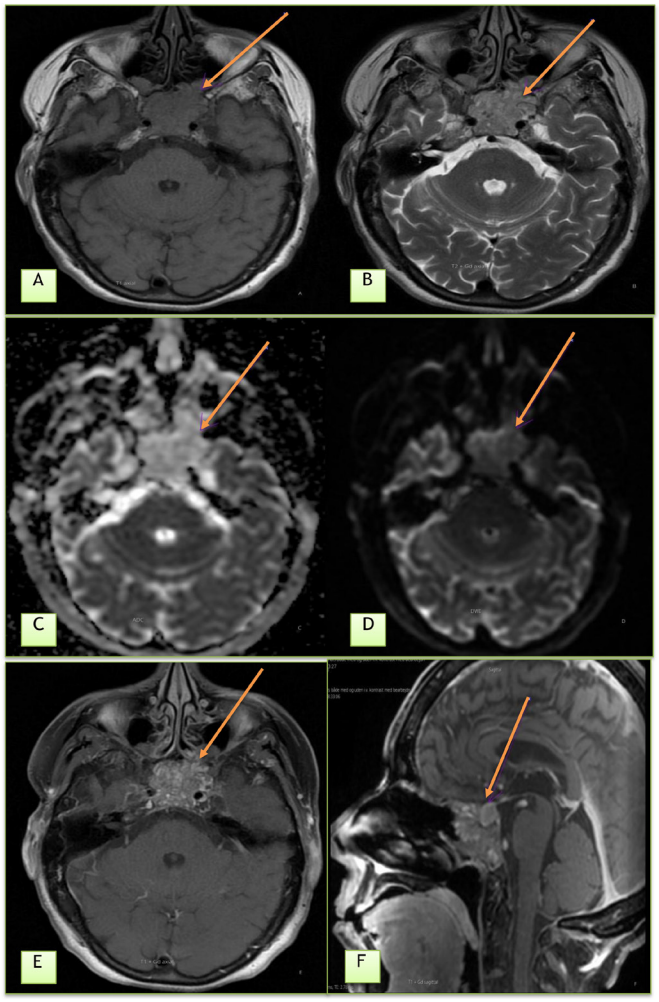

Figure 3 MR axial T1 without contrast shows an isointense tumour (A), axial T2 shows mild hyperintensity (B), DWI and ADC without diffusion restriction (C and D), axial and sagittal T1+gadolinium show moderate enhancement of the tumour (E and F) (Tumour has a grapelike structure that can easily be identified in T2 and T1+Gadolinium sequences).

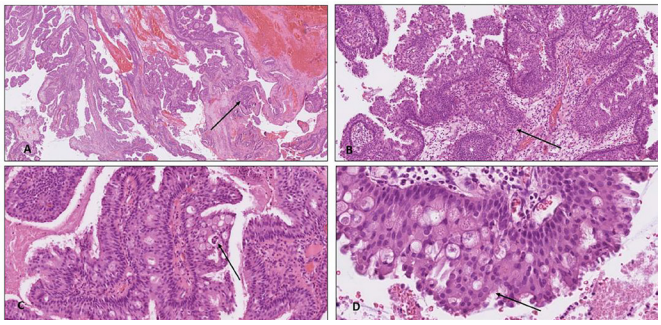

Figure 4 A low (A) and medium (B) power view of the tumour showing the predominantly exophytic and some endophytic (arrow) growth of multilayered epithelium. Medium-power view (C) shows mucous cells and small cavities, some with the microabscess structure of neutrophils (arrow). Mucous cells and cilia (arrow) are clearer on high magnification (D).

OUTCOME AND FOLLOW-UP

The patient's postoperative clinical course was uneventful, and she was sent home 3 days after the operation. The first postoperative follow-up was performed after 1 month, and the second follow-up was performed 6 months after the surgery. She did not have symptoms related to the OSP or complications of the surgery, and endoscopic examination of the paranasal sinuses showed that the patient is free of recurrence of OSP.

DISCUSSION

The OSP is the rarest type (3%–5%) of three morphologically distinct papillomas arising from the Schneiderian membrane of the nasal cavity and paranasal sinuses.[1–3] In 1991, the WHO classified sinonasal papillomas into three distinct histological subtypes: EP, IP and OSP.[4] The lateral wall of the nasal cavity, maxillary sinus and ethmoidal sinus involvement are the most reported locations of the OSP and in English literature, only a few cases with sphenoidal sinus involvement are reported.[13 18] Wang *et al* in 2022 reported six cases with OSP involving the sphenoidal sinus; although exclusive sphenoidal sinus origin was not specified.[19] In a retrospective study of 20 cases by Lilja *et al* in 2019, tumour location in the maxillary sinus (60%), nasal cavity including ethmoidal sinus (35%), frontal sinus (5%) and none in the sphenoidal sinus was observed.[11] Most of the patients presented in the fifth decade of life with no gender or race predominance.[3 4 8] Patients with OSP had complaints of unilateral nasal block, intermittent epistaxis, depreciating sense of smell and severe headache.[4 8 10] These lesions usually present as a fleshy pink, tan, red-brown or grey papillary or polypoid growth.[8 20] The tumour is characterised by recurrences (33%–40%),[3 12 20 21] aggressive local growth and significant malignant potential (4%–17%)[2 4 8 9] with invasive squamous cell carcinoma being the most frequently reported tumour.[3 4 6] Interestingly, our patient never had symptoms due to the sphenoidal sinus papilloma and was first highlighted with positive FDG uptake in bilateral sphenoidal sinus in a PET CT scan that was done with a suspicion of disseminated ovarian carcinoma in November 2023. Retrograde evaluation of previous CT scanning confirmed a comparatively smaller polypoid lesion in the left sphenoidal sinus. The treatment for OSP is complete surgical excision of the tumour. Postoperative chemotherapy and/or radiotherapy can be considered if there is malignant transformation of the lesion. Histologically, OSP demonstrates exophytic and/or endophytic (inverted) growth of multilayered epithelium, composed of columnar cells with abundant eosinophilic and granular cytoplasm. Nuclei are round to oval and can vary from vesicular to hyperchromatic. Intraepithelial mucus cells and mucus microcysts with neutrophils are quite common. The outer surface of epithelium may demonstrate cilia. Regular follow-up examinations should be done as OSP has a high recurrence rate, as mentioned above.[4]

Learning points

- Unilateral mucosal thickening of paranasal sinuses, even if it is an isolated sphenoidal sinus mucosal thickening with no existing symptoms, must also be given careful attention due to the chance of this process being an oncocytic papilloma with risk of malignant transformation.
- Sphenoidal sinus oncocytic sinonasal papilloma (OSP) can easily be misdiagnosed when there are nonspecific symptoms with an incidental finding of a polyp in the sphenoidal sinus, as OSP is a rare tumour and isolated sphenoidal sinus involvement is even rarer.
- Effective treatment involves complete removal of the tumour with wide endoscopic excision.
- Careful histological examination is needed to exclude the risk of malignant transformation.

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