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Case Series

Radiology Section

Computed Tomography and Magnetic Resonance Imaging of Preoperative Cases of Esthesioneuroblastoma (Olfactory Neuroblastoma): A Case Series

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ABSTRACT

Esthesioneuroblastoma is an uncommon aggressive tumour arising from the olfactory epithelium of superior nasal cavity, frequently invading the base of cranium, cranial vault and orbit. Contrast enhanced Computed Tomography (CT) and contrast enhanced Magnetic Resonance Imaging (MRI) were done for three patients which showed modified Kadish stage C tumour. Biopsy was done to confirm the diagnosis and patients were subjected to surgery and radiotherapy. The purpose of this case series was to report the radiological features of esthesioneuroblastoma. This is a report on three cases of different age groups who presented tumour with intracranial extension which were histopathologically proven as esthesioneuroblastoma.

Keywords: Chemotherapy, Nasal cavity, Radiotherapy, Surgery

INTRODUCTION

Esthesioneuroblastoma is also known as olfactory neuroblastoma (neuroectodermal tumour), these are uncommon malignant tumours originating in the olfactory epithelium in the superior nasal cavity of olfactory recess. Approximately, the incidence of esthesioneuroblastoma is 4 per 10,00,000 cases. A wide range of age groups can present with esthesioneuroblastoma and frequently in a bimodal distribution, occurring most frequently in the second and sixth decades. There is no racial predilection to esthesioneuroblastoma [1]. Commonly, males are seen affected more than females with esthesioneuroblastoma. The male to female ratio is approximately 1.2:1 [2-5].

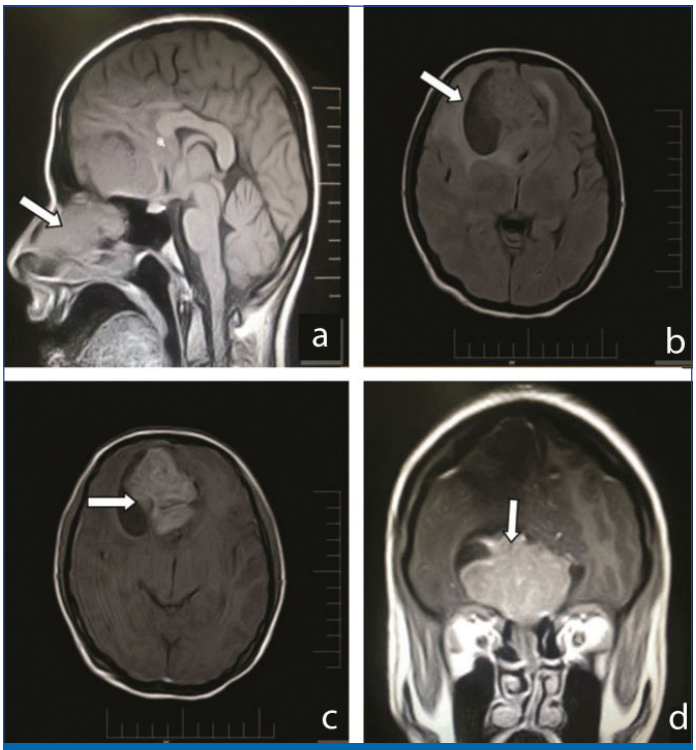
Most of the patients are locally diagnosed with advanced stages of the disease and hence, requires multimodality treatment in form of surgery, chemotherapy or/and radiotherapy.

CASE SERIES

Case 1

A young female in her third decade (27 years) presented with a mass in right nasal cavity, nasal bleeding and anosmia for a year. She underwent a lot of medications, but showed no improvement, recently she felt difficulty in breathing and severe frontal headache which were unresponsive to pain killers. On examination, a huge fleshy polypoidal mass was visible in right nasal cavity with non preservation of normal anatomy of nasal cavity and bleed with touch. Examination of the left nasal cavity, mouth, ears, neck and cranial nerves were within normal limited.

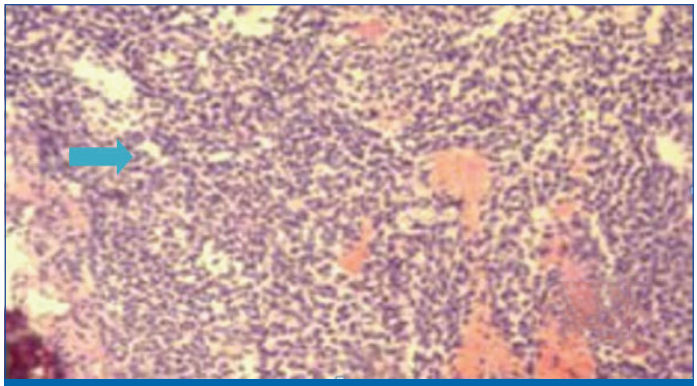
MRI with T1 axial and coronal sections, T2/FLAIR (Fluid Atenuated Inversion Recovery) axial sections and T1 post-gadolinium (contrast) axial and coronal sections were achieved. An isointense expansile mass lesion was noted in right nasal cavity with destruction of ethmoid air cells on T1WI [Table/Fig-1a] causing widening and destruction of right cribriform plate. FLAIR sequences showed significant perilesional oedema (hyperintensities), causing buckling and mass effect over the bilateral basi-frontal neuroparenchyma [Table/Fig-1b]. Postcontrast the lesion shows adjacent peritumoural cyst [Table/Fig-1c] with heterogeneous pattern of enhancement [Table/Fig-1d]. On Histopathological Examination (HPE) tumour cells were arranged in lobular pattern and large islands of small cells [Table/Fig-2].



[Table/Fig-1a-d]: a) The T1 MR saggital sequence shows expansile isointense mass lesion in right nasal cavity; b) Axial FLAIR sequences show significant perilesional oedema; c) T1+C axial section shows low intensity area (arrow) and peritumoural cyst; d) T1+C coronal section shows heterogeneously enhancing intracranial mass lesion.

Case 2

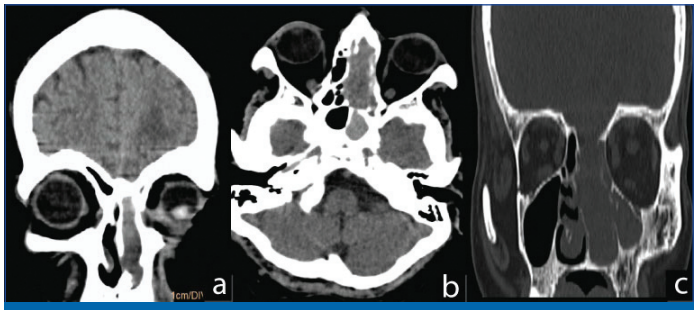
A 60-year-old male came to Ear, Nose and Throat (ENT) clinic and presented with history of headache, anosmia, episodic epistaxis, epiphora, left sided nasal blockage for a year and an episode of involuntary movements of both upper and lower limbs. He was



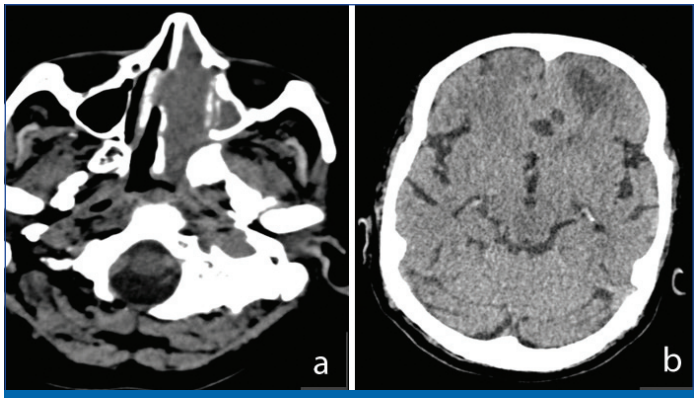
[Table/Fig-2]: Case-1, Microscopic section showing tumour cells arranged in lobular pattern and large islands of small cells (blue arrow). These cells are monotonous, having a hyperchromatic nucleus with inconspicuous nucleolus and mitotic activity (H&E, 100X magnification).

treated symptomatically for a year without any improvements. Recently, he developed balance disorder with an episode of seizure. On examination of nasal cavity, a pale colored soft fragile mass obliterating left nasal cavity bleed with touch. The assessment of sensory neuron and motor responses, especially reflexes were assessed and were unremarkable. Cold spatula test did not show fogging on left side. The patient was a known hypertensive and chronic smoker. Lab investigation such complete blood count, liver function test and renal function test were unremarkable.

CT of para-nasal sinuses and brain was performed with axial, coronal and saggital reformation which showed an expansile irregular soft tissue density mass lesion (Hounsfield unit 14 to 20) in left nasal cavity involving left maxillary and sphenoid sinus [Table/Fig-3a] causing obliteration of ostiomeatal complexes and extending superiorly intracranially into basi-frontal region crossing the midline. Destruction and thinning of left ethmoid air cells, left maxillary sinus walls, nasal septum, left inferolateral part of crista galli with widening and erosion of cribriform plate on left side [Table/Fig-3b,c] are extending posteriorly upto nasopharynx [Table/Fig-4a].

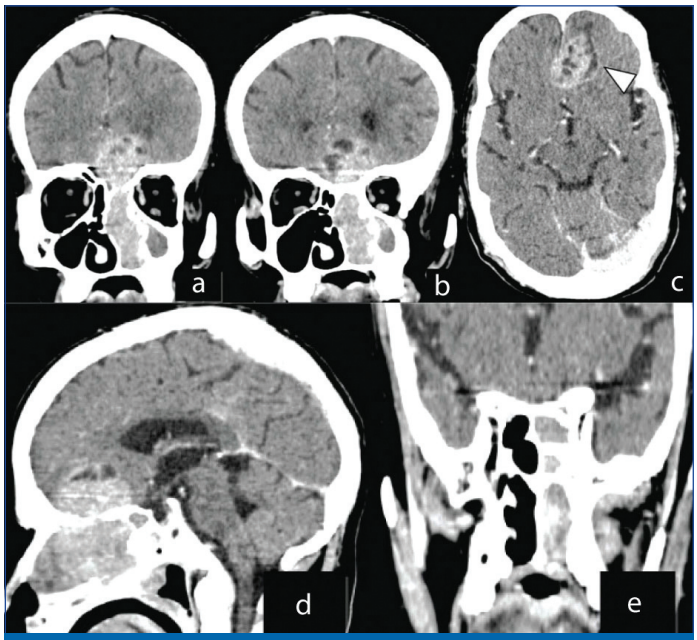


[Table/Fig-3a-c]: a) CT coronal section showing a heterogeneous mass lesion in left nasal cavity obliterating ostiomeatal complexes; b) CT axial section showing the mass lesion is diffusely infiltrating left maxillary and sphenoid sinuses; c) CT coronal section (bone window) showing mass lesion in diffusely infiltrating left para-nasal sinuses with widening and erosion of cribriform plate on left side.



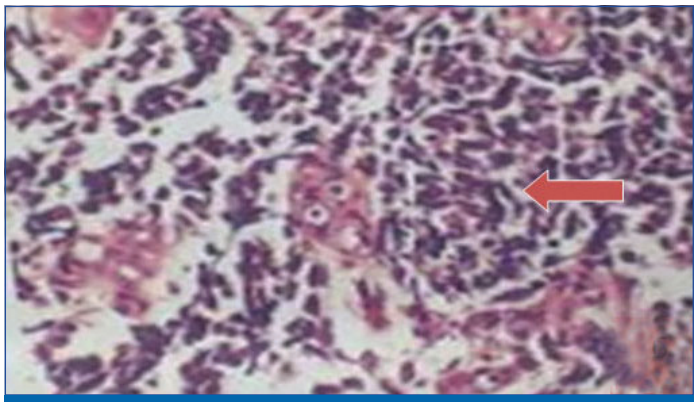
[Table/Fig-4a,b]: a) CT axial section showing heterogeneous mass lesion in nasal cavity extending upto nasopharynx; b) CT axial section at the level of thalamus showing a hypodense mass lesion in frontal region with few adjacent cysts.

On non contrast CT [Table/Fig-4b] there was a hypodense mass lesion with few cystic areas in frontal lobe in midline. On post-contrast (iohexol) CT, a large heterogeneously enhancing mass (Hounsfield unit 55 to 68) in left nasal cavity and sinuses [Table/Fig-5a,b] have seen extending intracranially in left frontal region crossing midline with adjacent peritumoural cyst ([Table/Fig-5c] arrowhead) with enhancing walls and significant peri-lesional oedema [Table/Fig-5d,e]. No evidence of intra-lesional calcifications/haemorrhage/intra-orbital extensions.



[Table/Fig-5a-e]: The heterogeneously enhancing mass lesion in left nasal cavity extending intracranially with peritumoural cyst (arrowhead).

On HPE examination, scant vesicular cytoplasm and scattered rosettes along with fibrillary matrix and mitotic activity with no evidence of necrosis was observed ([Table/Fig-6] red-arrow).

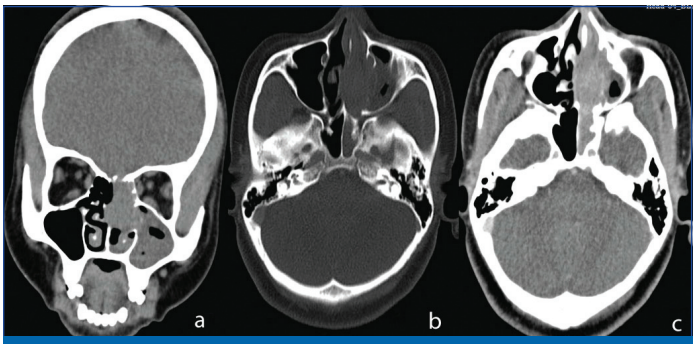


[Table/Fig-6]: Case-2, Microscopic sections showing tumour cells arranged in lobular pattern having a hyperchromatic nucleus with inconspicuous nucleolus, scant vesicular cytoplasm and scattered rosettes along with fibrillary matrix and mitotic activity with no evidence of necrosis (H&E, 400X magnification).

Case 3

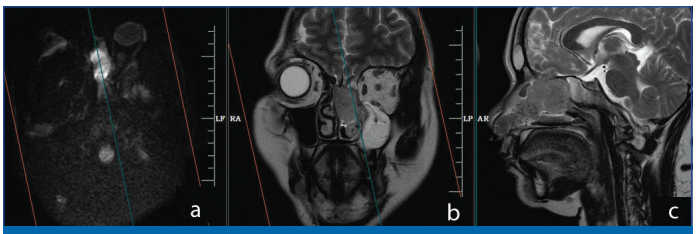
A 35-year-old female came with the complaints of nasal obstruction and increasing swelling over left cheek region and headache for a year. Recently, she developed severe recurrent epistaxis and complete anosmia and severe headache which was not relived with painkiller. No previous medical treatment or surgery was performed. On examination, a fleshy pinkish mass was visible in left nostril causing completely occupying left nasal cavity and firmed, non tender swelling with ill-defined margins over the left cheek and maxillary region and loss of smell sensation. There was no rise in local temperature in left cheek region. Cold spatula test did not show fogging on left side. Blood profile was within normal limits of complete blood count, liver function test and renal function test.

CT of para-nasal sinuses and brain was performed with axial, coronal and sagittal reformation which showed an ill-defined irregular heterogeneously enhancing mass lesion in left nasal cavity. The maxillary sinus completely obliterates left nasal cavity and extends to anterior cranial fossa through the cribriform plate. The lesion was causing destruction to medial wall of left maxillary sinus, uncinate process, superior, middle, inferior turbinate, nasal septum and superiorly cribriform plate [Table/Fig-7a-c].

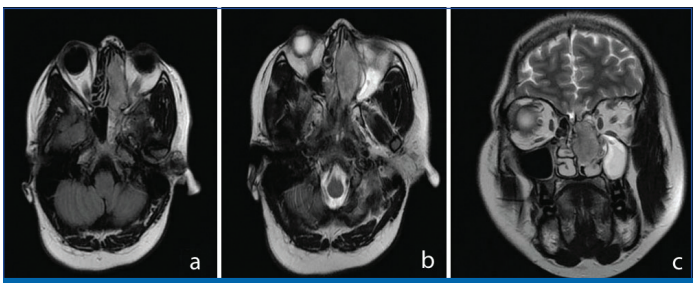


[Table/Fig-7a-c]: a) CT coronal section shows an isodense density mass lesion in left nasal cavity seen infiltrating left maxillary sinus; b) CT axial section (Bone window) shows a mass lesion in left nasal cavity seen infiltrating left maxillary and sphenoid sinus; c) Postcontrast CT axial section image shows that the tumour shows heterogeneous enhancement.

The heterogeneous mass lesion shows diffusion restriction on Diffusion Weighted Imaging (DWI) [Table/Fig-8a] with corresponding coronal and sagittal images [Table/Fig-8b,c]. MRI with T1 axial and coronal sections, T2/FLAIR, DWI, Apparent Diffusion Coefficient (ADC), Gradient Echo (GRE) axial sections and T1 post gadolinium (contrast) axial and coronal sections were achieved which showed an irregular heterogeneously enhancing mass lesion in left nasal cavity superiorly extending intracranially to anterior cranial fossa eroding cribriform plate on left side causing obliteration of fronto-ethmoidal recess and retention of secretions in left maxillary, sphenoid and frontal sinuses with downward and lateral displacement of left middle and inferior turbinates [Table/Fig-9,10].

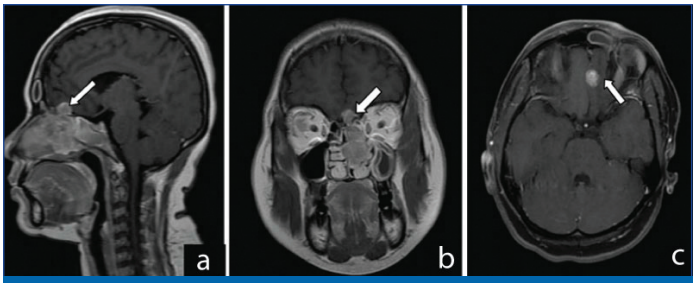


[Table/Fig-8a-c]: a) Axial section MR Diffusion Weighted Imaging (DWI) showing mild diffusion restriction; b & c) Coronal and sagittal section MR T2 weighted imaging showing a heterointense mass lesion in left nasal cavity, respectively.



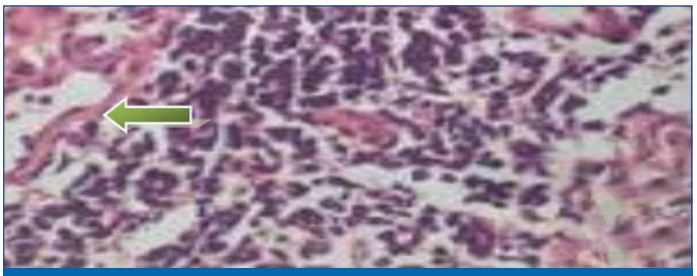
[Table/Fig-9a-c]: a,b) MR axial sections show a heterogeneous mass lesion in left nasal cavity causing obliteration of ostiomeatal complexes; c) MR coronal sections show heterogeneous mass lesion in left nasal cavity causing displacement of left middle and inferior turbinates.

A nasal cotton gauze soaked in a mixture of 5 mL of lidocaine 1% and 1 mL of adrenalin 1:50,000 is inserted in nasal cavity on the same side of lesion at an ear, nose and throat clinic then the biopsy specimen was taken under topical anesthesia. All the above three cases were diagnosed as esthesioneuroblastoma. HPE showed tumour cells arranged in lobular pattern and large islands of small cells, having



[Table/Fig-10a-c]: a,b) T1+C Saggital and Coronal sections showing a heterogeneously enhancing mass in left nasal cavity extending in intracranially; c) T1+C axial sections at midbrain level showing a enhancing mass lesion in anterior cranial fossa.

a hyperchromatic nucleus with inconspicuous nucleolus, [Table/Fig-11]. Differentials were olfactory neuroepithelioma and sinonasal carcinoma on imaging appears identical to esthesioneutoblastoma; however peritumoural cysts are uncommon in latter.



[Table/Fig-11]: Case-3, Microscopic sections showing tumour cells arranged in lobular pattern and large islands of small cells, having a hyperchromatic nucleus with inconspicuous nucleolus, scant vesicular cytoplasm, scattered rosettes along with fibrillary matrix and capillary dilatation (green arrow). No evidence of necrosis. Mitotic activity- present (H&E, 400X magnification).

DISCUSSION

Esthesioneuroblastoma is an unusual (1-5% of all nasal tumours) and destructive malignant neurogenic tumour located in the nasal cavity. Since, its initial description in 1924 by Berger and Luc [6] more than 1,000 cases of esthesioneuroblastoma were reported worldwide [7]. Symptoms are associated with the site of origin and extension/invasion of the tumour. Kadish S et al., 1976 accord tumour extensions as stage A, B, and C [8]. Later, Morita A et al., added stage D tumours as metastases in cervical lymph nodes and distant sites [9]. Biller HF et al., Dulguerov P and Calcaterra T are other two staging methods used [10,11]. The groups A-D are described below-

Group A: Mass lesion limited to the nasal cavity

Group B: Mass lesion limited to the nasal cavity and paranasal sinuses

Group C: Mass lesion extending beyond the nasal cavity and paranasal sinuses such as intracranial/intraorbital extension

Group D: Cervical nodal metastases

All three patients presented with Stage C in present study according to Kadish S et al., [8] with intracranial extension. The age group in present series showed patients ranged between 21 and 65 years old. There is still a lack of consensus over the incidence in both genders. Some reported that esthesioneuroblastoma affect male and female patients equally [12], some thought that it is slightly more common in males [13-16] or otherwise [17]. In general, the clinical symptoms of esthesioneuroblastoma were not specific and related to tumour sites and invasion [18]. The commonest presenting symptoms were unilateral nasal obstruction and anosmia in index study. Due to its vague symptoms and slow growing nature, esthesioneuroblastoma patients often have a long history of progressive symptoms for months, prior to diagnosis [11]. The average duration from symptom onset to management was 1 year in index study and six months (0–18 months) in the USA, reportedly [13]. This study established that MRI and CT manifestation of esthesioneuroblastoma are imprecise, sharing similar signal intensity and density features and

enhancement patterns with other nasal neoplasms [19,20]. The predominant site of primary tumour was the roof of nasal cavity in the region of the cribriform plate, laterally above the middle turbinate. Rare sites of initial incidence such as the sellar and parasellar region, nasopharynx, maxillary and sphenoid sinuses were not seen in this series [21-24]. Locally, esthesioneuroblastoma extended laterally and upwards to the paranasal sinuses or medially to the contralateral nasal cavity and into the cranial cavity or fossa orbitalis. Furthermore, esthesioneuroblastoma can also metastasise by lymphatic and haematogenic routes. It is reported that approximately 5% of patients have cervical lymph node metastasis with a cumulative cervical metastases rate of 25% or so [3,25] and approximately 10-30% of patients will develop distant metastases to the central nervous system (about 20-30%), lung, liver, skin, eye, bone, and parotid [20,26-30]. In present study, lymph nodes metastases were not seen. This report mainly concentrated on these three cases with direct intracranial extension and categorised them into three types based on the depth of invasion: brain parenchyma (Kadish Group C) (3/3), other types being in nasal cavity (Grade A) or para-nasal sinuses (Grade B). The brain parenchyma type had the primary tumour extend across the cribriform plate, growing through the dura and then infiltrate into the anterior frontal lobe. Even though the bulk of the tumour may lie intracranial, it is still attached to the cribriform plate.

Compared with the gray matter, the tumour parenchyma appears T2/FLAIR hyperintense and hypointense on T1-weighted images. FLAIR image exhibits the peritumoural oedema more clearly, but gadolinium-enhanced T1-weighted images visualise the tumoural boundary due to significant enhancement of all involved soft tissues. Peri-tumoural cysts (arrowhead in [Table/Fig-5c]) along the intracranial tumour margin are portrayed clearly. MR and CT images show that the tumour located at high in the nasal cavity extending through the left cribriform plate and invading the ipsilateral frontal lobe. Peri-tumoural cyst (arrows) along the margin of the intracranial tumour is portrayed clearly. Pre-contrast coronal CT image shows a hyperdense endonasal mass with slight high density to the surrounding soft tissue. Post-contrast coronal CT image shows the tumour enhances significantly and heterogenously.

No intra-lesional calcification can be considered pathognomonic for esthesioneuroblastoma. Presence of intracranial cysts in a sino-nasal mass with CNS extension is highly suggestive of esthesioneuroblastoma [31]. The base-of-skull type was revealed in three cases, where bulging duramater were demonstrated and thinned or destroyed cribriform complex were discovered during surgery. Due to the high aggressive behaviour and recurrence rate [28,32] of esthesioneuroblastoma, early diagnosis, accurate staging and close follow-ups are essential for optimistic patient management [33]. In these aspects, both MR and CT scanning are helpful in providing necessary information as to the tumour location and extension and proximity to the adjacent structures.

CONCLUSION(S)

Esthesioneuroblastomas presented as expansible intranasal lesions that unilaterally widens the olfactory recess. CT/MRI can help predict extent and staging of tumour, guide extent for biopsy, potentially helpful in choosing appropriate initial operative management.

This analysis of the case series of esthesioneuroblastomas suggests that intracranial extension and peritumoural cysts are more commoner and anosmia being the commonest symptom.

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