Original Research Paper

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MANAGEMENT OF RARE LESIONS AND MASSES OF NOSE AND PARANASAL SINUSES – APPROACHES AND MODALITIES

Praveena Varasala — Assistant Professor, Dept. of ENT, Rangaraya Medical College, Govt. General Hospital, Kakinada

Solomon Saawan Pudi — Junior Resident, Dept. of ENT, Rangaraya Medical College, Govt. General Hospital, Kakinada

Hemanth Ijju — Junior Resident, Dept. of ENT, Rangaraya Medical College, Govt. General Hospital, Kakinada

ABSTRACT Lesions of nose and paranasal sinuses are fairly common. The incidence of such lesions varies with climate, geographical area or with habits and occupational exposures in people. Treating such lesions varies with the stage of presentation and facilities available for treatment. Dept. of ENT, Govt. General Hospital, Kakinada is a tertiary care centre for the surrounding areas. A compilation of rare cases and modalities and approaches used to treat them have been made. Efficacy of treatment has been noted with each modality and approach.

KEYWORDS : . Teratocarcinosarcoma, Plasmacytoma, hemangiopericytoma, Rhinocerebral, Mucormycosis, meningioma, Ameloblastoma, Meningoencephalocoele, Rhinosporidiosis, Angiofibroma

AIM: Management of rare lesions of nose and paranasal sinuses by various surgical approaches

PATIENTS AND METHODS:

INCLUSION CRITERIA: All patients coming to casualty and ENT OPD, Govt. General Hospital, Kakinada between July 2014 and June 2017 with complaints of nasal mass, nasal obstruction and epistaxis have been evaluated and patients with evidence of rare lesions on clinical examination, pre op biopsy or CT scan have been included.

EXCLUSION CRITERIA: All patients unfit for surgery or patients choosing radiotherapy over surgery were excluded from the study.

METHOD OF COLLECTION OF DATA: This is a prospective study carried out in the Dept. of ENT, Govt. General Hospital, Kakinada between July 2014 and June 2017. The study includes 14 cases of rare sino-nasal masses and lesions. The study was approved by the ethics committee of the institution. All cases had been first evaluated clinically. Nasal endoscopy and imaging studies were done wherever applicable. Biopsy was taken in few select cases preoperatively in OT setting and all cases post operatively for histopathological diagnosis and confirmation. Immunohistochemistry was done wherever required for diagnosis. Details of type of lesion, appearance at presentation, clinical, radiological and histopath ological findings were recorded and data was analysed. Cases were treated both by endoscopic approach and by open approaches assisted endoscopically wherever possible and sent for radiotherapy post operatively whenever required and followed up for variable periods and treatment results were analyzed.

OBSERVATIONS AND RESULTS:

14 patients with rare lesions were treated.

Table 1: Sex distribution of cases:

TOTAL PATIENTS	MALE	FEMALE
14	7	7

7 of the 14 patients were male [50%] and 6 patients [50%] were female.

Table 2: Age distribution of cases:

AGE	10-20	21-30	31-40	41-50	51-60	61-70
PATIENTS	2	3	1	4	2	2

Most patients are in 41-50 years age group [35%].

Table 3: Diagnosis and treatment modality

DIAGNOSIS	NO. OF CASES	TREATMENT APPROACH
Teratocarcinosarcoma	1	Lateral Rhinotomy
Hemangiopericytoma	1	Endoscopic removal
Angiofibroma	2	Endoscopic removal
Nasal Meningioma	1	Mid Facial degloving + medial maxillectomy
Plasmacytoma	1	Weber Ferguson+ medial maxillectomy
Mucormycosis	1	Weber Ferguson-U/L Maxillectomy
Mucormycosis	1	Endoscopic excision
Ameloblastoma	1	Endoscopic excision
Fibrous dysplasia frontal sinus	1	Endoscopic removal
Sphenoid sinus carcinoma	1	Endoscopic removal
Adenoid cystic carcinoma	1	Lateral rhinotomy + partial maxillectomy
Meningoencephalocoele	1	Endoscopic closure of leak
Rhinosporidiosis	1	Endoscopic removal with cautery of base

Open approaches were attempted in extensive lesions and in malignancies. Endoscopic removal was attempted in situations where lesion was amenable to endoscopic approach or at the end of open approaches to confirm complete disease clearance.

Table 4: Post operative Radiotherapy

DIAGNOSIS	NO. OF CASES	Period
Teratocarcinosarcoma	1	Refused radiation therapy
Hemangiopericytoma	1	33 gray. 2 gray/fraction
Plasmacytoma	1	33 gray. 2gray/fraction
Sphenoid sinus carcinoma	1	35 gray. 2 gray/fraction
Adenoid cystic carcinoma	1	27 gray. 2 gray/ fraction

Patients were sent for radiotherapy 2 weeks after the surgery and followed up after radiotherapy and treated for radiation side effects.

Post operative radiotherapy was given in the form of external beam radiotherapy with multi leaf collimators using teleCobalt60 machine.

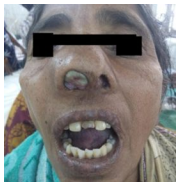
Table 5: Recurrence

DIAGNOSIS	NO. OF CASES	TREATMENT MODALITY
Teratocarcinosarcoma	1	Refused radiotherapy

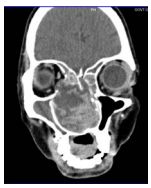
Only case with a recurrence was a patient with sinonasal teratocarcinosarcoma

Individual case reports:

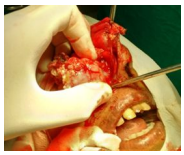
1. Sinonasal teratocarcinosarcoma: A 62year old female presented with H/O right sided nasal obstruction for 1 year with decreased perception of smell on right side with epistaxis and epiphora since 1 month. She also had foul smelling purulent nasal discharge, chronic head ache with loss of weight and appetite. She had H/O exposure to smoke while cooking in firewood stove. She had no exposure to wood dust or industrial chemicals. On examination she had an ulceroproliferative growth in right nasal cavity with hemorrhagic spots, covered with mucopurulent pus with deviation of septum to left. Biopsy of the lesion preoperatively suggested poorly differentiated squamous cell carcinoma. Pre-operative CECT showed heterogenous enhancing mass in nasal cavity, maxillary sinus and nasopharynx with invasion of lamina and involvement of conal and extra conal compartments. Moure's lateral rhinotomy was done and mass was removed entirely. Attachments were found on septum anteriorly. There were hemorrhagic and necrotic areas apart from a globular mass which filled entire cavity which pushed the septum contralaterally. Endoscope assisted removal was done to clear the mass from posterior ethmoid sinuses. Turbinates were eroded. Excised tissue biopsy revealed teratocarcinosarcoma. Immunohistochemistry confirmed the diagnosis.



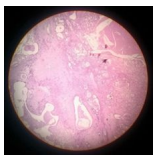
Pre Op patient photo



Pre op CECT coronal view



Intra Op picture

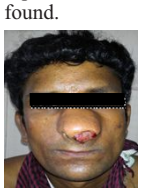


Histopathology slide

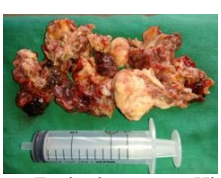
Patient and her attendants were explained about nature and prognosis of disease and advised radiotherapy. Patient did not consent for radiotherapy due to difficulty in follow up. She had locoregional recurrence from septum 6 months later and died 14 months after surgery.

2. Extra medullary Plasmacytoma: A 24 year old male patient presented with history of left side nasal obstruction, ipsilateral decreased perception of smell and visible nasal mass for the last two years which was slowly increasing in size with nasal deformity and pain in nose since 2 months (Secondary infection and bone erosion result in pain.)He had similar complaint 3 years back for which he was operated [at a different centre] and was symptom free for 12 months. On HRCT PNS, there was a homogenously enhancing soft tissue opacity seen occupying the left nasal cavity, left maxillary antrum, left ethmoids and sphenoid sinus causing deviation of nasal septum with evidence of destruction. Preoperative biopsy was suggestive of inflammatory polyposis and was inconclusive. Weber Ferguson approach was taken and mass was cleared from nasal cavity, sinuses and choana. Anterior face of maxilla was drilled for approach to maxillary sinus and nasal cavity. Attachments were from lateral wall and ethmoid cells. Excised mass was fleshy, yellow-pink to dark-red, pedunculated mass with an uneven, non-ulcerated surface.

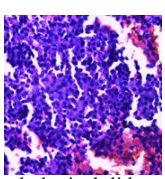
Biopsy was suggestive of extramedullary plasmacytoma and Immuno histochemistry concluded the diagnosis. He was tested for bony involvement and metastasis by blood smears, xrays, bone marrow aspiration and biopsy and ultrasonography of abdomen and none was found.



Pre Op patient photo



Excised mass



Histopathological slide

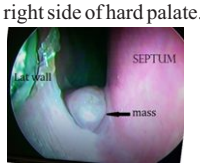
Patient underwent post-operative radiotherapy. Patient feels normal 18 months after surgery and has returned to work and is on follow up nasal endoscopy once in 3 months, radiographs and ultrasound scans once in 6 months.

3. Ameloblastoma: A 47 year old female came with dental pain of 1 month duration with loosening of teeth and foul breath. On examination, she had bony swelling of ipsilateral hard palate adjacent to affected loose teeth. She underwent surgery for the same diagnosis 8 years ago by Calwel Luc approach. Pre operative biopsy was suggestive of Ameloblastoma. CT was suggestive of bony growth. On nasal endoscopy, there was a fistula between the lesion in palate and nasal floor. At surgery the bony lesion was drilled and removed via nasal cavity with assistance of endoscope. Bone was removed till normal bone was seen. The affected teeth were extracted and scope was passed from the openings created in gums into maxillary antrum and into the area of lesion to check for complete removal. The gums were sutured and floor of nasal cavity was left to epithelialise and heal secondarily. Patient had discharge from the fistula in nose for about 2 months [Patient was diabetic on insulin therapy] after which all symptoms subsided. She is on follow up for 18 months so far.

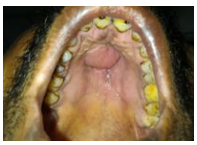
4. Fibrous dysplasia of frontal sinus: A 27 year old woman presented with slowly progressive protrusion of left eye ball since 4 months. On examination the eye ball was pushed laterally, inferiorly and proptosed with increased inter pupillary distance. CT revealed nonenhancing lesion in frontal sinus. Draff IIb procedure was done and lesion was removed totally with help of 70 degree endoscopes and angled drills and malleable probes. Bone was removed upto periosteum of globe. Biopsy reported fibrous dysplasia. Patient is on follow up for 11 months so far and has had gradual resolution of proptosis.

5. Hemangiopericytoma: A 40 year old hypertensive male presented with nasal obstruction and epistaxis since 1 month. On nasal endoscopy, mass was pink and fleshy and was bleeding on touch. Biopsy [done at different centre] revealed hemangiopericytoma. CECT showed enhancing areas in left nasal cavity without evidence of bone destruction. Surgery was endoscopic with blood loss around 200ml. Bleeding stopped as soon as the whole mass was removed. Bleeding was controlled by suction cautery and pressure packing. Patient underwent post operative radiotherapy and has been disease free for 10 months.

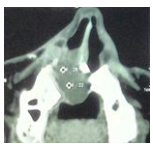
6. Nasal meningioma: A 46 year old male agricultural labourer presented with history of right nasal obstruction and epistaxis. He was a smoker and alcoholic with no exposure to industrial chemicals and fumes or wood dust. Nasal endoscopy revealed soft, pink polypoidal fleshy mass in right nasal cavity, not bleeding on touch and probe couldn't be passed laterally. There was a hard smooth bulge over the right side of hard palate.



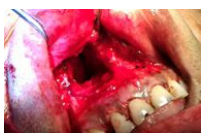
Polypoidal nasal growth



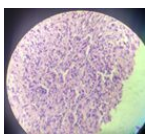
Palatal swelling



CT with nasopalatine mass



Mid facial degloving intra op



Histopathology slide

This was the 4th procedure on the patient as the lesion had recurred 6 months apart since 2.5 years. He had earlier undergone endoscopic removal, Caldwel Luc and endoscopic approaches in order. At the time of 4th procedure, he had involvement of palate and lateral wall for which a midfacial degloving approach was undertaken and procedure included medial maxillectomy with use of burrs to remove lesion from frontonasal process of maxilla and from floor of nasal cavity. Patient developed an oronasal fistula for which a dentist was consulted and a palatal prosthesis was designed and mucosa of palate and floor of nasal cavity grew above it. Patient has been on follow up and disease free for a year now.

7. Squamous cell carcinoma of sphenoid sinus: A 65 year old

hypertensive male presented with total blindness in both eyes. He had history of epistaxis which he neglected for a long

time. CECT scan revealed enhancing mass lesion involving sphenoid sinus and proximity to optic nerve and carotid arteries. Endoscopic excision was done with use of neurosurgical drills, bone rongeurs and ball probes to remove bone for delineation of optic nerves and carotid arteries. Post operative high dose radiation was given. Patient recovered upto perception of light in both eyes. Patient is on follow up for 10 months after surgery.

8. Angiofibroma: A 18 year old male presented with nasal obstruction since 2 months and epistaxis. Nasal endoscopy revealed a globular reddish pink mass. CT scan revealed enhancing mass in nasal cavity, nasopharynx and pterygopalatine fossa. Endoscope assisted excision of mass was done with suction cautery and pressure packing to control bleeding. Turbinates were removed for better exposure. Bone rongeurs were used for exposure of pterygopalatine fossa and mass was removed with suction cautery. Blood loss was 400ml. Patient is on follow up for 2.5 years without recurrence.

9. Angiofibroma: A 13 year old male presented with progressive nasal obstruction, snoring and epistaxis. Mass on endoscopy was reddish pink and patient had frog face deformity. CECT showed mass in nasal cavity, nasopharynx and pterygopalatine fossa. Endoscope assisted excision consisted of medial maxillectomy and removal of medial part of posterior wall of maxillary sinus using drill and bone rongeurs. Mass was removed with bipolar and monopolar suction cautery and mass delivered from nasopharynx via oral cavity. Blood loss was 200ml. Child is on follow up for 1.5 year after surgery without recurrence.

10. Meningoencephalocoele with CSF rhinorrhoea: A 42 year old female presented with watery nasal discharge of 2 months duration. She had no history of trauma and T2 MRI revealed meningo encephalocoele herniating through cribriform plate and CSF leak. Surgery was endoscopic with removal of middle turbinate and cautery of prolapsed meninges. The area of leak was inspected. Autologous fat and composite graft middle turbinate were used to seal the leak. Surgicel was placed over the graft site and Merocel was used to pack the nasal cavity. Patient was put on mannitol and acetazolamide post operatively and advised against straining. Patient in on follow up for 10 months without recurrence of CSF leak.

11. Mucormycosis with poorly differentiated squamous cell carcinoma: A 47 year old female presented with epistaxis of 2 weeks duration. She had cheek swelling and loss of sensations over it. Patient also had medial rectus palsy. She had history of exposure to smoke while using firewood stove for cooking. Pre operative biopsy was suggestive of poorly differentiated squamous cell carcinoma. CECT revealed enhancing lesion of maxilla and ethmoids. Consent was taken for maxillectomy. Weber Ferguson approach was used and osteotomies were made and maxilla was removed entirely ipsilaterally. Bleeding was controlled by cautery. Prosthesis was created with acrylate and inserted for support of facial skin. Nasogastric feeding was done for 1 week. Obturator was designed and inserted 1 week post operatively after which patient resumed semisolid food. Medial rectus palsy subsided and ocular movements became normal.

Biopsy revealed coexistent mucormycosis along with squamous cell carcinoma and patient was sent for MRI which suggested rhinocerebral mucormycosis with involvement of carotids and temporal lobes. She was put on amphotericin and monitored till improvement. Patient developed a CVA 2 months postoperatively and passed away.

12. Mucormycosis: A 56 year old female with diabetes and hypertension presented with epistaxis and nasal obstruction since 2 months. Nasal endoscopy revealed ethmoidal polyps and biopsy was suggestive of inflammatory polyposis. CT scan suggested changes consistent with fungal involvement. Surgery was endoscopic with removal of all polyps from sinuses and septa of sinuses were found to be hard and had gritty consistency while removal. The operating surgeon had a clinical diagnosis of chondrosarcoma on palpation. Bone curettes were used to scrape the unhealthy bone. Post-operative histopathological examination revealed mucormycosis. Patient was put on amphotericin. She is on follow up for 18 months without recurrence.

13. Adenoid cystic carcinoma: A 50 year old female agricultural labourer who uses firewood for cooking presented with left sided epistaxis, loosening of teeth and left cheek swelling with loss of sensations over it. Pre operative biopsy revealed adenoid cystic carcinoma and CT showed enhancing lesion of left hard palate and maxillary sinus. Moure's lateral rhinotomy and medial maxillectomy were done and anterior face of maxilla was drilled and lesion removed. Partial maxillectomy and palatectomy was done to remove the part of palate involved. Biopsy confirmed adenoid cystic carcinoma. No metastasis was found. Patient was sent to post op radiotherapy and has recovered from radiation mucositis and is on follow up for 14 months without recurrence.

14. Rhinosporidiosis: A 21 year old male student presented with epistaxis. He is from Srikakulam, Andhra Pradesh and has habit of swimming in ponds. On closer endoscopic examination the mass resembled rhinosporidiosis. Endoscopic removal of mass was done with cauterization of base from floor of nasal cavity and nasopharynx. Larynx was inspected for lesions and none were found. He was put on post-operative dapsone for 3months. He is on follow up for 10 months without recurrence.

Discussion:

Sino nasal teratocarcinosarcoma (SNTCS) is a very rare and aggressive malignant neoplasm histologically characterized by the combination of one or many components of epithelial and mesenchymal elements[1].. Multimodality treatment, in the form of a combination of surgery, radiation therapy, and chemotherapy, appears to be the optimal approach[2]A combination of radiotherapy and surgical treatment offers the best five-year survival rate (50%); followed by only surgical treatment (47%). In the recurrent or metastasis lesion, adjuvant chemotherapy may improve the survival rate since the metastatic tissue often contains sarcomata components.

Extra medullary plasmacytoma is a rare neoplasm characterized by monoclonal proliferation of plasma cells. Most lesions occur in the head and neck, primarily in the upper aerodigestive tract. The nasal cavity and nasal septum are the most common sites of occurrence.[3] Extramedullary plasmacytoma (EMP) represents approximately 3% of all plasma cell neoplasms[4]. The treatment of EP is surgical resection and radiotherapy[5]. Chemotherapy may be considered for patients with refractory or relapsed disease[6]. Follow-up radiological and serum electrophoresis is required after treatment to detect recurrences and progression to myeloma. Approximately 10% of EP has multiple sites of involvement[7]. The rate of progression to multiple myeloma is lower than in solitary bone plasmacytoma (SBP), ranging from 11% to 30% and is associated with a poorer prognosis[7].

Ameloblastoma is a benign odontogenic tumor generally present in the jaw bone. The tumor originates from the residual epithelium of the tooth germ, epithelium of odontogenic cysts stratified squamous epithelium and epithelium of the enamel organ. It represents approximately 1% of oral tumors. About 80% of ameloblastomas occur in the mandible mainly the third molar region and the remaining 20% in the upper jaw. Ameloblastoma clinically appears as an aggressive odontogenic tumor, often asymptomatic and slow-growing, with no evidence of swelling[8]. Ameloblastoma has a high rate of local recurrence if it is not adequately removed. Radical surgical resection of ameloblastoma is the treatment of choice[9].

Fibrous dysplasia is a rare and indolent benign tumor of bone. Although it is a benign tumor, it has malignant transformation potential. Fibrous dysplasia can involve a single or multiple bones. The maxilla and mandible are the most common sites in the head and neck region. Paranasal sinus involvement is rare. Surgical excision is the preferred treatment of patients with symptomatic fibrous dysplasia[10] Hemangiopericytoma behaves aggressively, demonstrating greater than 50% local recurrence and 10% metastatic disease. Extended surgical resection is traditionally considered the most effective therapy for all HPC[11]

Approximately 6–17 % of all meningiomas can be found in extracranial or extra spinal sites. The head and neck is the most frequent location of extraneuraxial meningiomas, commonly seen as an extension of primary intracranial tumor (secondary extracranial meningioma). Less than 2 % of all meningiomas arise at a primary ectopic site (primary extracranial meningioma)[12] Treatment of primary extracranial meningioma is surgical excision because meningiomas

are known to be radio resistant and recurrence is very rare following adequate surgical resection[13].

Headache and visual disturbances are common symptoms of sphen oidal sinus tumors. The presence of cranial neuropathies, although common, suggests a less successful outcome. It is advantageous if the surgical team has had experience with both open and endoscopic approaches to the sphenoidal sinus. Patient outcome is maximized by the use of aggressive multidisciplinary therapy.[14]

Treatment of nasopharyngeal angiofibroma includes lateral rhinot omy, transpalatal, transmaxillary, or sphenoethmoidal route is for small tumors (Fisch stage I or II).The infratemporal fossa approach is used when the tumor has a large lateral extension. The midfacial degloving approach, with or without a Le Fort osteotomy, improves posterior access to the tumor[15]. The facial translocation approach is combined with Weber-Ferguson incision and coronal extension for a frontotemporal craniotomy with midface osteotomies for access. An extended anterior subcranial approach facilitates en bloc tumour removal, optic nerve decompression, and exposure of the cavernous sinus.Some authors advocate the use of intranasal endoscopic surgery for lesions with limited extension to the infratemporal fossa. Image-guided, endoscopic, laser-assisted removal has also recently been used[16].

Endoscopic approaches for CSF rhinorrhoea when compared with external techniques have several advantages, including better field visualization with enhanced illumination and magnified as well as angled visualization. Another advantage is the ability to more accurately position the underlay or overlay grafts. Multiple studies demonstrate a 90-95% success rate with closure of skull base defects using the endoscopic approach. [17]

Treatment of rhinocerebral mucormycosis includes reversing underlying immunocompromised states, administering systemic antifungals, performing urgent surgical debridement. Correcting hypoxia, acidosis, hyperglycemia, and electrolyte abnormalities is critical to the successful management of this condition. Discon tinuation or maximally reducing chemotherapy and immunos uppressive therapy is desirable if clinically possible. Any steroid medication, antimetabolites, or immunosuppressants that the patient is on should also be addressed and discontinued if appropriate. [18]

Adenoid Cystic Carcinomas are common in minor salivary glands about the mouth, uncommon in parotids and rare in Nose and para nasal Sinuses. Histologically they are composed of small deeply basophilic nuclei with characteristic gland like structures. Adenoid Cystic Carcinoma of Nose and para nasal Sinuses has a unique natural history. They most commonly present with mass or epistaxis. Even though slow growing, they have a propensity for frequent local recurrence and early perineural and haematogenous spread[19]. Lungs are the most common site for metastasis. Lung metastasis are usually multiple and prolonged survival with multiple metastasis is not unusual. Treatment is surgical removal with radiotherapy[20].

Nasal rhinosporidiosis may present with unilateral nasal obstruction or epistaxis. Other symptoms may include local pruritus, coryza with sneezing, rhinorrhea, and postnasal discharge (drip) with cough. Patients often report a sensation that a foreign body is present in their nasal canal. Rhinosporidiosis is treated with surgical excision and multiple reports of successful treatment of individuals with long courses of dapsone have been published. [21,22]

Conclusion:

Most lesions and masses of the nose and para nasal sinuses can be treated endoscopically but some extensive lesions require more radical open approaches with post operative radiotherapy. Radiation mucositis largely defines a patient's rehabilitation and most are rehabilitated in a year after therapy. Availability of equipment like rongeurs, drills and endoscopes with good knowledge of anatomy and approaches plays a big role in effective removal of lesions.

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