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Original Research Article

A clinicopathological study of the lesions of paranasal sinuses causing proptosis

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ABSTRACT

Background: The orbit and its contents lie in close proximity to the paranasal sinuses. The aim of the study was to analyse the lesions of paranasal sinuses causing proptosis.
Methods: All patients with proptosis secondary to paranasal sinus disease were taken up for a systematic otolaryngological and ophthalmological clinical evaluation and investigation using a standard proforma. Incidence of various disease of paranasal sinuses causing proptosis, age and sex distribution, incidence of different symptoms and signs and its association with proptosis were estimated. The reversible and irreversible nature of the proptosis in relation to the degree of proptosis and the histopathological nature of the disease were studied.
Results: Malignant diseases were found to be the commonest lesion that caused proptosis. Squamous cell carcinoma was the most common histopathological type. Invasive fungal sinusitis among inflammatory and inverted papilloma among benign tumours were the commonest lesions that cause proptosis. Male preponderance was seen in the inflammatory and malignant lesions and the commonest symptoms were headache and nasal obstruction. Malignancies caused faster proptosis than benign and inflammatory lesions. Analysis of X-ray and computed tomography (CT) findings were also done.
Conclusions: Malignant lesions are the most common para nasal sinus disease that cause proptosis. Duration of illness and degree of proptosis varied with the underlying disease process. With inflammatory proptosis some underlying sinus pathology in addition to the sinusitis is found in all cases. CT is a better modality to identify underlying pathology.

Keywords: Headache, Paranasal sinus, Proptosis, Squamous cell carcinoma

INTRODUCTION

Progressive proptosis is a perplexing situation for the clinician and an ominous sign for the patient. There are few more perplexing problems in clinical medicine than gradually progressive unilateral exophthalmos. The evaluation and differential diagnosis is difficult and demands close cooperation among various specialists.[1] Most ENT surgeons unless specifically trained view the orbit and its contents with trepidation.

The orbit represents an important anatomic cross road of central nervous system, nose and paranasal sinuses, the face and structures concerned with the support and functions of the eye. It is intimately related to paranasal sinuses on its three sides. It is separated only by thin plates of bones from the sinuses located around it. In addition, apart from the anatomical continuity, the veins of the orbit are in direct communication with the veins of face, nose, paranasal sinuses, pterygoid plexuses and cavernous sinuses. The fact that none of these veins is

provided with effective valve, make the orbit the meeting place of surrounding pathological disturbances.

Orbital extension of infection and neoplasia of the paranasal sinuses may occur through natural pathways like open suture lines, congenital dehiscence in the orbital wall and foramina for nerves and vessels, defects in the bony walls due to fractures or necrosis by acute infection or erosion by chronic disease or retrograde thrombophlebitis. Because of these factors orbital involvement is not uncommon in diseases of the nose and paranasal sinuses. It heralds, in many situations grave prognosis or increased morbidity. Hence its early detection merits great importance in otolaryngology practice.

The orbit is composed of 7 bones. The lateral wall is formed by the greater wing of the sphenoid apically and the frontal and zygomatic bones facially. The floor is formed from the sphenoid, the orbital process of the palatine bone, and the orbital process of the maxillary bone. The medial wall is formed from the lesser wing of the sphenoid, the ethmoid bone, the lacrimal bone, and the frontal process of the maxilla. The roof of the orbit is derived from the sphenoid and the frontal bones.[2] Any demand for increased accommodation within it either by edematous fluid, blood, inflammatory infiltrate or neoplastic growth or any encroachment from outside by swelling around it can only be met by pushing forward of the eyeball, otherwise called proptosis. The average volume of the orbit is 30 ml and 70% of which is occupied in normal individual by retrobulbar and peribulbar structures.[3] Orbit being a fixed bony cavity, an increase of orbital content by 4 ml produce almost 6 mm of proptosis. Hence proptosis is one of the common and early manifestations of orbital involvement. On many occasions it may be the only presenting symptom or sign of an underlying paranasal sinus disease. The directions of the proptosis may help one to locate the site of lesion in this, otherwise clinically in-accessible region. Therefore detecting and understanding the proptosis is imperative for effective dealing of paranasal sinus diseases so that further damage to the orbital content can be prevented. Unusual cases of bilateral proptosis due to orbital hemorrhage caused by factor IX deficiency (hemophilia B) has also been reported.[4]

Proptosis is defined as any forward displacement or bulging and is frequently used to describe the abnormal protrusion of the eyeball from its normal position in the orbit. The term exophthalmos is more specific to the organ involved. These two terms are used synonymously, but the term exophthalmos is more commonly used to denote proptosis of endocrine or metabolic origin. Except in advanced stages of disease, paranasal sinus lesions seldom cause bilateral proptosis.

In the present study, it was observed that patients with proptosis of paranasal sinus origin reporting to the out-patient department directly or referred by the ophthalmologist. Florid proptosis will not be missed by anyone but for detection of mild early proptosis which is clinically more important, the clinician should be aware of its existence and is trained to detect it. The dictum that the eyes will see only what the mind knows, holds good here.

The aims of the study were to assess the prevalence and to evaluate the natural history of diseases of paranasal sinuses causing proptosis, identify the early proptosis associated with para-nasal sinuses, to have an understanding about the pathogenesis and pathological changes involved in the process and to document the reversible and irreversible changes in the orbit with regard to duration and degree of illness.

METHODS

The study was done on patients who attended ENT out-patient department of DM Wayanad Institute of Medical Sciences from July 2013 to April 2020. All patients who had proptosis secondary to the paranasal sinus were included in the study. Cases having proptosis due to other ENT disease like nasopharyngeal lesions were not included. An attempt was also made to note the general incidence of proptosis with ENT diseases. No attempt is made to take the incidence of proptosis in general as only small percentage of proptosis cases presented with ENT disease and majority were managed by Ophthalmology Department. A total number of thirty patients were evaluated in the study.

All patients with proptosis secondary to paranasal sinus disease were taken up for systematic otorhino-laryngological and ophthalmological clinical evaluation and investigation using a standard proforma. Efforts were made to quantify the proptosis during the initial presentation and during follow up, using Luedde's exophthalmometer. Only essential investigations were done since in most of the cases computed tomographic study were done. Only in those cases when the diagnosis were obvious by clinical examination that computed tomography (CT) was avoided. In necessary cases histopathological diagnosis were made prior to definite treatment.

The evaluation of the results were made keeping in mind the aims of the study. Efforts were made to identify the early proptosis by clinical examination and quantify the degree of proptosis in relation to the duration of symptoms. Incidence of various disease of paranasal sinuses causing proptosis, incidence according to age and sex, incidence of different symptoms and signs and its association with proptosis in these cases were estimated. The reversible and irreversible nature of the proptosis in relation to the degree of proptosis and the histopathological nature of the disease were studied. Importance of early detection of proptosis to prevent further orbital damage was emphasized. All patients were reviewed for follow up for 2 weeks after the discharge

and afterwards at monthly interval for next 3 months and thereafter at 6th, 9th, 12th months for a period of 1 year and thereafter to report if there are any symptoms.

Ethical approval

All procedures in studies involving human participants were in accordance with the ethical standards of the national research committee and with the 1964 Helksinki declaration and its later amendments or comparable ethical standards.

Treatment

Treatment policy in each case was individualized depending on the nature of the disease whether benign or malignant and degree of orbital involvement. Medical, surgical or radiotherapeutic methods were used either as single modality or in combinations.

RESULTS

Out of the total 30 subjects, majority belonged to age group of 41-50 years and the least number of patients were in 61-70 years age group. The average age of patient with malignancy was 51 years, the range being 7-60 years. Those with mucocele have an average age of 49 years, the range being 22-72 years. The patients with inflammatory disease have an average age of 33 years distributed between 5-50 years. Only one child was seen in this study. Age of patients with benign tumor varied from 22-45, average being 39. That of fibrous dysplasia from 14-33 years with average being 22 years. Traumatic cases were 19 and 25 years old.

Males accounted for the majority number of patients in this study amounting to 20 (67%) and females were 10 (33%).

Table 1: Analysis of the chief symptoms.

Symptoms	No. of cases	Percentage
Headache	26	87
Nasal obstruction	20	67
Nasal discharge	18	60
Bulging of the eye	18	60
Watering from the eye	17	51
Facial swelling	12	40
Bleeding from the nose	7	23
Defective vision	6	20
Swelling of the eyelids	5	17

Inflammatory and malignant conditions were predominantly seen in males. Occupation of these patients varied from manual laborers to skilled workers. No wood industry worker had been noted. In our study we noted a male preponderance in inflammatory, malignant lesions and fibrous dysplasia whereas female preponderance in mucocele. Equal sex distribution was seen among benign tumors and traumatic cases.

Table 2: Presentation of physical signs.

Sign	No. of cases	Percentage
Proptosis	30	100
Limitation of eyeball movement	14	47
Epiphora	17	57
Decreased visual acuity	10	33
Destruction or bulging of orbital wall	22	73
Telecanthus	11	37
Periorbital and facial swelling	12	40
Abnormal nasal findings	28	93

It is seen that commonest complaint was headache and nasal obstruction. Only 60% of these patients complained of bulging of their eye (Table 1).

From the Table 1 and 2 it is evident that only 60% of patients with clinically evident proptosis, noticed and complained of bulging of their eyes. 40% were not aware of its presence.

Table 3: Incidence of various diseases of paranasal sinuses causing proptosis.

Name of disease	No. of cases	Percentage
Malignancy of paranasal sinuses	8	27
Mucocele/pyocele	7	23
Acute and chronic sinusitis	6	20
Benign tumors	4	13
Fibrous dysplasia	3	10
Traumatic	2	07
Total	30	100

Table 4: Inflammatory lesions that caused proptosis.

Inflammatory lesions	No. of cases	Percentage
Acute maxillary sinusitis	1	16.6
Infected dental cyst maxilla	1	16.6
Diabetic gangrene maxilla	1	16.6
Acute ethmoiditis with subperiosteal abscess	1	16.6
Invasive fungal sinusitis	2	33.4

Analysis of number of patients presenting with varying degree of proptosis showed 21 (70%) cases in mild (20-23 mm), 8 (27%) in moderate (24-28 mm) and 1 (3%) in severe (29 mm and above) degree of proptosis. Two cases

from moderate group become severe degree during follow up. One was malignancy of the frontal sinus and other malignancy of the maxilla.

Malignant diseases are found to be the commonest paranasal sinus lesion that causes proptosis. Mucocele comes second. All of the seven causes of mucoceles were of frontal or ethmoid origin. They caused proptosis directed downward and outward. Of this one case was pyocele with osteitis of the frontal sinus floor (Table 3).

Table 5: Analysis of relation between duration of illness and degree of proptosis.

Disease	Mild	Moderate	Severe
Inflammatory	1.5 months (6)	-	-
Malignancy	4.6 months (4)	3 months (3)	3 months (1)
Benign tumour	27 months (2)	42 months (2)	-
Traumatic	1 day (1)	3 months (1)	-
Mucocele	32 months (5)	42 months (2)	-
Fibrous dysplasia	58 months (3)	-	-
Total	21	8	1

Relation between duration of illness and degree of proptosis varies with disease entity. Average duration is given in months and number of cases in each group are given on parenthesis. Inflammatory conditions were of two types, a chronic variety presented as fungal sinusitis and the acute sinusitis causing orbital cellulitis. Acute cases were of less than 1 week duration and chronic variety were of 3-4 months duration. In all these conditions proptosis were of mild degree. Malignant lesions does not show a linear relationship between the duration and progression of proptosis. It appears to depend on the invasive nature of the tumour. Squamous cell carcinoma shows relentless speedy progression once it involved the orbit. Esthesioneuroblastoma shows a relatively slower pace. Benign tumours have a linear relationship with duration and degree of proptosis and duration varied from two to six years. Traumatic cases presented dramatically with proptosis of sudden onset. In one case secondary to intranasal ethmoidectomy, proptosis was noted immediately after the surgery. The other of accidental origin had proptosis secondary to formation of carotid cavernous fistula. Proptosis developed 3 months after the accident. It was pulsatile and rapidly progressive with chemosis and lid edema. Patient had severe recurrent epistaxis. Fibrous dysplasia was characterized by very slow progression of proptosis and that too of mild degree. Duration varied from one and a half years to eight years. It has been noted that right orbit was involved in 16 cases and left in 14 cases. Inflammatory lesions were found distributed equally on both sides. 2 cases seen in a child and adolescent were on left side. Malignancies were distributed equally on both sides. Benign tumours were seen more on the right side. Proptosis due to surgical trauma occurred on right side.

Table 6: Incidence of benign tumours.

Benign tumours	No. of cases	Percentage
Inverted papilloma	2	50
Neurofibroma ethmoid	1	25
Paraganglioma of maxillary sinus	1	25
Total	4	100

Inverted papilloma is the commonest benign tumours of the paranasal sinuses that cause proptosis. Para ganglioma of the maxillary sinus is noted as a very rare tumor in this series. Three cases of fibrous dysplasia was noted. Two were of monostotic type involving the frontal and ethmoid respectively, and one polyosteotic type. Two cases of proptosis due to trauma to paranasal sinuses were noted. One developed acute right proptosis following intranasal ethmoidectomy and another due to carotid cavernous fistula secondary to fracture of sphenoid sinus in a craniofacial injury. This carotid cavernous fistula was characterized by pulsatile proptosis. Even though there was loud bruit heard over the eyeball and skull, subjective tinnitus was significantly absent. His hearing was normal in both ears.

Table 7: Involvement of various paranasal sinuses by malignancy.

Sinus	No. of cases	Percentage
Maxillary	3	38
Ethmoidal	4	50
Frontal	1	12
Total	8	100

Ethmoidal sinus malignancy was mostly involved in the causation of proptosis followed by maxillary sinus.

Table 8: Histopathological types of these tumors.

Types	No. of cases	Percentage
Squamous cell carcinoma	4	50
Adenoid cystic carcinoma (maxilla)	1	12
Esthesioneuroblastoma	2	25
Small cell carcinoma	1	12

Squamous cell carcinoma was the most common histopathological type of malignancy followed by esthesioneuroblastoma (Table 8).

Opacity of the sinus followed by destruction of the sinus wall were the commonest findings on X-ray (Table 9).

Table 9: Analysis of plain X-ray study of paranasal sinuses (n=30 cases).

Findings	Benign (n=22)	Malignant (n=8)
	N (%)	N (%)
Opacity of the sinus or sinuses	22 (100)	8 (100)
Destruction of the sinus wall	11 (50)	7 (88)
Bilateral involvement	4 (18)	None
Sclerosis of bone	3 (14)	-
Fracture of sinus wall	1 (5)	-

Table 10: Analysis of CT findings (n=20 cases).

Radiological findings	Benign (n=13)	Malignant (n=7)
	N (%)	N (%)
Involvement of sinus cavities	13 (100)	7 (100)
Destruction of sinus wall	9 (69)	7 (100)
Orbital involvement	13 (100)	7 (100)
Intracranial involvement	2 (15)	4 (57)
Nasopharyngeal extension	2 (15)	4 (57)
Bilateral involvement	4 (31)	-
Contrast enhancement	2 (15)	4 (57)

Except one, all malignant cases had CT evaluation and in all these cases involvement of sinuses, sinus wall and the orbit were seen. Plain X-ray failed to show bone destruction in 12% of malignancy. Plain X-ray study and CT evaluation demonstrate that benign lesions also could cause bone destruction. By CT it is found difficult to demonstrate whether the soft tissue shadow in the sinus cavities and nasopharynx were due to invasion by the tumor or inflammation or edema. In the case of carotid cavernous fistula, CT evaluation could pinpoint the diagnosis, but it could not differentiate the associated venous congestion and edema of submucosa of nasopharynx and sinus cavities from an expanding soft tissue tumor. Fibrous dysplasia on plain X-ray and CT had ground glass appearance due to sclerosis of the bone. The orbit is an area of considerable importance to the ENT surgeon. It may be involved in diseases of nose and paranasal sinuses or surgical procedure can involve this area intentionally or accidently. Proptosis is the cardinal sign of orbital involvement. Thirty patient presenting with proptosis secondary to disease of paranasal sinuses were studied. Malignancy was found to be the most common cause followed by mucocele. Rare cases like neurofibroma of the ethmoid, paraganglioma of the maxillary sinus and carotid cavernous fistula were also seen in this series. Only sixty percentage of patient were aware of their proptosis. Destruction or distention of the orbital wall was seen in 73% of patient and 93% had abnormal nasal findings. 70% had mild proptosis and the degree of proptosis was indicating of the degree of orbital involvement. In benign lesions the periorbita were found intact where as in all operated cases of malignant tumors periorbita was found invaded.

DISCUSSION

The importance of orbit to the ENT surgeon lies more in its anatomical position than in its contents. Disease arising from the paranasal sinuses which surround it on three sides easily invade the orbit. Orbit behaves as a closed box comparable to cranium. It is bounded behind and on four sides by rigid bony walls and in front by the eyeball and septum orbitale. Any demand for increased accommodation with in it, can only be met by pushing forward of the eyeball, otherwise called the proptosis.

In this study consisting of 30 patients with proptosis secondary to diseases of the paranasal sinuses, neoplastic lesions of sinuses were found to be the commonest cause forming 40% of the lesions. Mucocele with an incidence of 23% form the second most common cause which is followed by inflammatory lesion (20%) fibrous dysplasia (10%) and traumatic lesions making 7%. There has been wide disagreement as to the frequency of the various lesions chiefly due to the means used in collecting the data. Palmer et al found that diseases of paranasal sinuses caused 3-10% of unilateral proptosis, 15% caused by vascular and 70% by primary orbital tumors. According to him mucocele is the most common disease of the sinuses which causes proptosis. Williamson-Noble et al stated inflammatory diseases of the paranasal sinuses as the commonest cause of ocular and orbital complication and he found proptosis as one of the common signs.[5] Advent of newer antibiotics, improved medical care and increased health awareness of the people have changed the incidence of sinus infection in the community and drastically reduced its complication. We found only 6 cases (20%) caused by sinusitis. Schramm et al in a series of 302 patients with orbital cellulitis found 80% of these patient has associated paranasal sinus disease.[6] Watters et al found these to be 84%.[7]

Henderson et al reporting on mayo clinic series of 764 cases of orbital tumors found 343 cases (45%) were secondary orbital tumor.[8] In our study neoplastic lesion form 40% of all sinonasal lesions that caused proptosis. Of these 67% (8 cases) are malignant and 33% (4 cases) are benign. Johnson et al reporting on 79 cases of sinus tumors found 49 cases of orbital involvement (66%).[9] 68% of them were malignant and 32% were benign. These incidence tallies well with our series. Mohan et al reporting on 31 cases of paranasal sinuses tumor involving the orbit found that there were 21 (70%) malignant and 10 (30%) benign tumors.[10] This agrees well with our findings.

Inverted papilloma, commonest benign tumor in this series, formed 6.6% of the sinus lesions causing proptosis. Johnson et al reported a slightly higher incidence than ours. Although osteoma of the paranasal sinus is considered to be common benign tumor of the sinuses by Attalah et al.[11] We did not come across any case.

We had one case of neurofibroma of the ethmoid a rare entity in the paranasal sinus region. Harrison reviewing 639 cases of nasal tumors found only seven neurofibromas.[12] Present series is too small to compare with that of Sir Harrison. Tallbote reports only 12 such cases in the literature, we had one case in a female aged 22 year with moderate proptosis.[13]

Holt et al in series of 727 patients with facial injuries reported ocular involvement in 18% of cases.[14] We did not come across any facio-maxillary trauma that produced proptosis directly due to orbital injury.

We had a case of acute proptosis of right eye, developed following functional endoscopic sinus surgery. Freedman et al reviewing 1000 consecutive intranasal ethmidectomies at Mayo clinic, reported only 4 cases of orbital hematoma and proptosis following surgery.[15] Friedman et al reviewing 1300 cases reported similarly.[16]

In 1955 Lamber cited by Bordley et al described the frontal sinus mucocele as the most common nasal condition to produce eye symptoms.[17] In Henderson's series 8.5% were mucocele and Shields reported that 2% of all orbital tumors were mucocele.[18] We found that mucocele is the second commonest condition to produce proptosis. We had three cases of fibrous dysplasia constituting 10% of the sinus lesions causing proptosis.

In our series patients with inflammatory diseases has an average age of 33 years distributed between 5-50 years. Only one child and adolescent were seen. This finding is in great variance with findings of other author. Schramm et al, Fearon et al, Welsh et al and Morgan et al found 50-80% of their patients with orbital sepsis were children.[6,19-21]

The low incidence of children in the present series may be due to the fact that availability of better medical care and wide use of newer antibiotics would have resulted in early control of upper respiratory infection. The second factor may be that all children having orbital complications may not be undergoing investigations to rule out the underlying sinus infections and may not be attending the otolaryngology department. Third factor may be that those adults who are having some underlying basic pathology of the sinuses, may be having persistent sinus infections and more prone for orbital complications as seen in our study. Four of the six patients in our series had such factors. Two had fungal sinusitis of ethmoids which was eroding the lamina papyracea. One had infected dental cysts of the maxilla and the other had uncontrolled diabetes mellitus and associated vasculitis and gangrene of the maxilla. These findings indicate that orbital infection spreading from the sinus in an adult should make one suspect some underlying pathology in addition to sinusitis. We did not seen any case of inverted papilloma in children even though Eavey reported five cases in children.[22]

Study of sex distribution in this series show a male predominance of 67%. Helliwell et al reported anaplastic carcinoma of glandular origin were more common in women with 64% involving the orbit.[23]

Occupation of these patients varied from manual labour to skilled work. The one patient who had adenoid cystic carcinoma of the maxillary sinus was not employed in wood industry. Klintberg et al reported that adeno carcinoma of the sinuses has a predilection for patients with wood dust exposure especially furniture workers.[24]

CONCLUSION

Malignant neoplastic lesions are the most common para nasal sinus disease that cause proptosis. The fact that only sixty percentage of patients with proptosis were aware of its presence and seventy percentage of them are having only mild proptosis emphasize the need for a conscious effort on the part of clinician to detect early proptosis. The relationship between duration of illness and degree of proptosis varied with the underlying disease process. Malignant lesions do not show a linear relationship between the duration of illness and the degree of proptosis. It appears to depend up on the invasive nature of tumor. Among malignant tumors those arising from the maxillary sinus are found to cause greater degree of proptosis since the orbit is accessible from three sides. In all benign conditions, irrespective of the degree of proptosis, the periorbita remained intact. Whereas in malignant lesions the periorbital was found invaded and penetrated. The presence of proptosis is an indication of the advanced stage of the underlying paranasal sinus disease. In adults with inflammatory proptosis some underlying sinus pathology in addition to the sinusitis is found in all cases. In orbital cellulitis, the proptosis persisted for longer period of time even after control of infection, probably due to slow resolution of underlying thrombophlebitis. In orbital cellulitis, an axial proptosis turning in to eccentric one, denotes development of sub periosteal abscess and the direction of proptosis indicate site of abscess. The frontal mucocele is commonly associated with ethmoid mucocele, together forming a single sac and the orbital involvement is almost always through the lamina papyracea rather than the floor of the frontal sinus.

The CT scan is found to be the ideal investigation for accurate assessment of sino-orbital lesions, even though it often fails to differentiate inflammatory changes from expanding soft tissue tumors. Regression of proptosis following treatment occurs in all benign lesions with in a

relatively short period, where as in malignant lesions the proptosis progresses to severe degree or necessitate orbital exenteration.

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