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# TABLE OF CONTENTS

<table>
<thead>
<tr>
<th>Content page:</th>
<th>Page #</th>
</tr>
</thead>
<tbody>
<tr>
<td>Thyroid Disorders in Childhood and Adolescence: Retrospective Assessment of Clinical Data in a Nigerian Teaching Hospital. Alphonsus N. Onyiriuka, Phillip O. Abiodun and Louis C. Onyiriuka:</td>
<td>3 – 11</td>
</tr>
</tbody>
</table>

## CASE REPORTS

<table>
<thead>
<tr>
<th>Title</th>
<th>Page</th>
</tr>
</thead>
<tbody>
<tr>
<td>Progressive Hemifacial Atrophy- A Case Report: Sumona Pal and Shruthi Hegde:</td>
<td>35 - 41</td>
</tr>
<tr>
<td>Oral Myiasis – A Case Report: Jyothi S Kumar:</td>
<td>47 – 50</td>
</tr>
<tr>
<td>Instructions to Authors:</td>
<td>57 – 62</td>
</tr>
</tbody>
</table>
THYROID DISORDERS IN CHILDHOOD AND ADOLESCENCE: RETROSPECTIVE ASSESSMENT OF CLINICAL DATA IN A NIGERIAN TEACHING HOSPITAL.

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ABSTRACT:
In Nigeria, thyroid disorders in childhood and adolescence have not been sufficiently studied. Where studies are available, they were either conducted decades ago or they involved only adults. The objectives are to describe the pattern of thyroid disorders among children and adolescents seen in a Nigerian teaching hospital and highlight the management challenges encountered. In this retrospective study, the case notes of all the children and adolescents with thyroid disorders seen in the Paediatric Endocrine-Metabolic Clinic and of those admitted into the wards of the University of Benin Teaching Hospital (UBTH) from 2005 to 2011 were audited. The total number of cases seen at the paediatric clinics of the Department of Child Health, UBTH was derived from the clinic attendance register of the department. Of the 8,350 cases seen during the period, 9(0.11%) had thyroid disorders, representing one per 930 cases. Of the 9 patients with thyroid disorders, 6(66.7%) had hyperthyroidism, 2(22.2%) had nongoitrous hypothyroidism and one (11.1%) had euthyroid goiter. The overall mean age at presentation for thyroid disorders was 11.2±4.3 years (95% Confidence Interval, CI = 8.4-14.0) and female-to-male ratio was 4:1. For the patients with hyperthyroidism, the mean age of presentation was 12.8±3.1 years (95% CI= 10.3-15.3) and female-to-male ratio was 5:1. The two children (a boy and a girl) with nongoitrous hypothyroidism were aged 3.5 and 6.0 years respectively. The mean duration of symptoms before presentation was thyroid disorders 1.72±1.2 years (95% CI=0.94-2.50), hyperthyroidism 8.5±1.5 months (95% CI=7.3-9.7). The only case of euthyroid goiter (female) presented at the age of 14 years. The two children with nongoitrous hypothyroidism had florid signs of hypothyroidism, such as growth retardation, mental retardation and delayed developmental milestones at presentation. A high clinic default rate was observed. Hyperthyroidism was the most common form of thyroid disorder observed and patients with thyroid disorders tended to present late.

Key words: adolescence, childhood, pattern, thyroid disorders.
(Submitted December 2011, Accepted April 2012)
INTRODUCTION:
Thyroid disorders is one of the endocrine disorders commonly encountered in childhood and adolescence and they manifest with qualitative or quantitative alterations in thyroid hormone secretion, goiter or both [1,2]. Insufficient hormone secretion results in hypothyroidism and excessive secretion cause hyperthyroidism or thyrotoxicosis. Sometimes goiter exists with normal thyroid function. Although thyroid dysfunction causes disturbances of metabolic function in both children and adults, the effect on cognitive function, growth and development are unique to children [1]. Despite this well recognized profound effect of thyroid dysfunction in children, there are very few studies that have examined childhood and adolescent thyroid disorders in Nigeria and other developing countries [3]. Majority of the available data either involved only adults or were conducted decades ago [3-6]. The aetiology, prevalence, clinical presentation, and clinical course of thyroid disorders in children and adolescents substantially differ from that of adults [7]. The incidence of thyroid disorders in children and adolescents appears to be increasing [8,9]. For instance, in the TennCare cohort of children without Down syndrome, an increase of 26% in rate of medically treated thyroid disease was observed when 2002-2003 was compared to 1995-1997 [9]. The study was based on prescription database. Laditan et al, [3] reported that 37.0% of children with endocrine disorders seen at the University College Hospital (UCH), Ibadan, Nigeria over a five-year period had thyroid disorders. In that report, late presentation was emphasized. In most parts of Africa and other resource-poor countries, the diagnosis of thyroid disorders depends largely on clinical acumen supported by laboratory tests [3]. Some studies have separately concluded that the pattern of thyroid disorders in a given population was dependent on its iodine-intake status [10-13]. Given that iodine-intake status varies between populations, one may surmise that the pattern of thyroid disorders may differ; justifying the study of pattern of thyroid disorders among children and adolescents in Nigeria.

The purpose of this study was to describe the clinical pattern of thyroid disorders in children and adolescents seen in the Department of Child Health, University of Benin Teaching Hospital (UBTH), Benin City Nigeria, between 2005 and 2011 and highlight some of the management challenges encountered.

SUBJECTS AND METHODS:
The study was conducted in the Department of Child Health, UBTH, Benin City, Nigeria and it involved patients seen between 2005 and 2011 which came mainly from Edo state and the neighbouring states of Delta, Ondo and Kogi. The Paediatric Endocrine-Metabolic Clinic (PEMC) of UBTH receives referrals from both
within and outside the hospital (UBTH). The study was approved by the hospital authority.

In this retrospective study, the cases were identified by examining the relevant hospital attendance and admission registers and auditing the case notes of children seen at the PEMC and of those admitted into the paediatric wards (UBTH). The diagnosis of thyroid disorders was based on clinical features and thyroid function tests which included determination of serum levels of thyroxine (T4), triiodothyronine (T3) and thyroid-stimulating hormone (TSH). Information extracted included age, gender, clinical features, duration of symptoms before presentation, laboratory tests results, management challenges, and outcome of admission. The total number of cases seen in the paediatric clinics of the Department of Child Health, UBTH was noted. Data analysis involved calculation of percentages, means, ratios and confidence intervals.

RESULTS:

During the 7-year period under review, a total of 8,350 cases were seen at the Paediatric Consulting Clinic of the Department of Child Health, UBTH. Of these number, 9(0.11%) had thyroid disorders, representing about 1 per 930 cases. Forty nine (0.6%) patients with endocrine disorders was seen during this period and 9(18.4%) had thyroid disorders. Of the 9 patients with thyroid disorders, 6(66.7%) had hyperthyroidism, 2(22.2%) had nongoitrous hypothyroidism and one (11.1%) had euthyroid goiter. The mean age at presentation of thyroid disorders was 11.2±4.3 years (95% Confidence Interval, CI=8.4-14.0) and female-to-male ratio was 4:1. For patients with hyperthyroidism, the mean age at presentation was 12.8±3.1years (95% CI=10.3-15.3) and female-to-male ratio was 5:1. The mean duration of symptoms before presentation for all the thyroid disorders combined was 1.72±1.2 years (95% CI= 0.94-2.50) while for hyperthyroidism it was 8.5±1.5 months (95% CI= 7.3-9.7). The mean duration of symptoms before presentation in the two children with nongoitrous hypothyroidism was 4.75±2.18 years (95%CI=1.74-7.76). The 6-year old boy with hypothyroidism had globally delayed developmental milestones: crawled after age of 12 months, walked at about age of 3 years but unable to run at 6 years of age. Started talking at the age of 4 years but words are not clearly pronounced. The patient was in KG 1 at the age of 7 years; can say “2” and “A” but can neither count nor recite alphabets. Height was 101cm at 6 years of age. He was the only male child out of four children in the family. The mother admitted she did not notice the child’s problems on time and that when she eventually did, she had hoped the child will overcome the developmental challenges with time. The 14-year-old girl who presented with goiter was found to be euthyroid and physical examination did not reveal any abnormality. She achieved menarche at the age of 13 years. There was no positive family history of thyroid
disorders in any of the patients. The presenting complaint in both patients with hypothyroidism was “Does not play/dull/sluggish.” Physical examination revealed that both patients had growth retardation, facial puffiness, mental retardation and delayed language development (Table 1). No history of use of oral contraceptive. Examination of the goiter revealed a diffuse, enlarged, soft gland with smooth skin and a bruit. As shown in Figure 1, goiter, lid retraction/stare, weight loss and tachycardia were present in all the six patients with hyperthyroidism. The 7-year old boy with nongoitrous hypothyroidism had short stature (height 101 cm). The two patients with hypothyroidism had low serum levels of T4 and T3 with elevated TSH whereas the patients with hyperthyroidism had elevated serum levels of T4 and T3 with either low or normal TSH levels (Table 2). Clinic attendance was generally poor except for one of the children with hypothyroidism. Of the six patients with hyperthyroidism, only one still attends follow up clinic. The mother of one of the adolescents with hyperthyroidism refused to come and pick up her daughter from the hospital for over 2 months after discharge, necessitating employing the services of law enforcement agents to repatriate the child to the parents.

Table 1: Age and gender distribution of eight children with thyroid disorders

<table>
<thead>
<tr>
<th>Patient</th>
<th>Thyroid disorders</th>
<th>Age at presentation (Years)</th>
<th>Gender</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Nongoitrous hypothyroidism</td>
<td>3.5</td>
<td>F</td>
</tr>
<tr>
<td>2</td>
<td>Nongoitrous hypothyroidism</td>
<td>6</td>
<td>M</td>
</tr>
<tr>
<td>3</td>
<td>Hyperthyroidism</td>
<td>7</td>
<td>M</td>
</tr>
<tr>
<td>4</td>
<td>Hyperthyroidism</td>
<td>12</td>
<td>F</td>
</tr>
<tr>
<td>5</td>
<td>Hyperthyroidism</td>
<td>12</td>
<td>F</td>
</tr>
<tr>
<td>6</td>
<td>Hyperthyroidism</td>
<td>14</td>
<td>F</td>
</tr>
<tr>
<td>7</td>
<td>Hyperthyroidism</td>
<td>15</td>
<td>F</td>
</tr>
<tr>
<td>8</td>
<td>Hyperthyroidism</td>
<td>17</td>
<td>F</td>
</tr>
<tr>
<td>9</td>
<td>Euthyroid goiter</td>
<td>14</td>
<td>F</td>
</tr>
</tbody>
</table>
Table 2: Clinical features seen in two children with hypothyroidism

<table>
<thead>
<tr>
<th>Clinical features</th>
<th>Patient 1</th>
<th>Patient 2</th>
</tr>
</thead>
<tbody>
<tr>
<td>“Does not play/dull/sluggish”</td>
<td>Positive</td>
<td>Positive</td>
</tr>
<tr>
<td>Gender</td>
<td>Male</td>
<td>Female</td>
</tr>
<tr>
<td>Age at presentation</td>
<td>6 years</td>
<td>3.5 years</td>
</tr>
<tr>
<td>Facial puffiness</td>
<td>Positive</td>
<td>Positive</td>
</tr>
<tr>
<td>Growth retardation</td>
<td>Positive</td>
<td>Positive</td>
</tr>
<tr>
<td>Mental retardation</td>
<td>Positive</td>
<td>Positive</td>
</tr>
<tr>
<td>Language</td>
<td>Delayed</td>
<td>Delayed</td>
</tr>
<tr>
<td>Weight</td>
<td>14 kg</td>
<td>10.5 kg</td>
</tr>
<tr>
<td>Height</td>
<td>101 cm</td>
<td>83 cm</td>
</tr>
<tr>
<td>Upper segment: Lower segment ratio</td>
<td>Increased</td>
<td>Increased</td>
</tr>
<tr>
<td>Bone age retardation</td>
<td>Positive</td>
<td>Positive</td>
</tr>
<tr>
<td>Muscle hypertrophy</td>
<td>Negative</td>
<td>Negative</td>
</tr>
<tr>
<td>Goiter</td>
<td>Negative</td>
<td>Negative</td>
</tr>
</tbody>
</table>

Table 3: Serum levels of thyroxine (T4), triiodothyronine (T3), and thyroid stimulating hormone (TSH) according to diagnosis in nine children with thyroid disorders.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Thyroid disorder</th>
<th>T4 (mcg/dl)</th>
<th>T3 (ng/ml)</th>
<th>TSH (mU/ml)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Non-goitrous hypothyroidism</td>
<td>2.6</td>
<td>0.5</td>
<td>38.5</td>
</tr>
<tr>
<td>2</td>
<td>Non-goitrous hypothyroidism</td>
<td>1.5</td>
<td>0.6</td>
<td>50.7</td>
</tr>
<tr>
<td>3</td>
<td>Hyperthyroidism</td>
<td>12.6</td>
<td>5.2</td>
<td>4.1</td>
</tr>
<tr>
<td>4</td>
<td>Hyperthyroidism</td>
<td>12.8</td>
<td>6.0</td>
<td>4.2</td>
</tr>
<tr>
<td>5</td>
<td>Hyperthyroidism</td>
<td>15.7</td>
<td>6.2</td>
<td>3.2</td>
</tr>
<tr>
<td>6</td>
<td>Hyperthyroidism</td>
<td>16.5</td>
<td>6.8</td>
<td>3.3</td>
</tr>
<tr>
<td>7</td>
<td>Hyperthyroidism</td>
<td>17.4</td>
<td>7.0</td>
<td>3.1</td>
</tr>
<tr>
<td>8</td>
<td>Hyperthyroidism</td>
<td>19.8</td>
<td>7.6</td>
<td>3.4</td>
</tr>
<tr>
<td>9</td>
<td>Euthyroid goiter</td>
<td>7.6</td>
<td>1.8</td>
<td>4.5</td>
</tr>
<tr>
<td></td>
<td>Normal values in UBTH laboratory</td>
<td>4.8-10.8</td>
<td>0.7-2.0</td>
<td>0.4-6.2</td>
</tr>
</tbody>
</table>
Figure 1: Presenting features seen in six patients with hyperthyroidism

DISCUSSION:
In the present study the prevalence of thyroid disorders was 1.7 fold higher than the prevalence (0.07%) reported, three decades ago, from UCH, Ibadan Nigeria [3]. There is no readily available explanation for the higher prevalence. The prevalence of thyroid disorders is higher in girls than boys with a ratio of 4:1. This is not surprising as other studies have reported a similar female preponderance [3,8,14]. In the present study, hyperthyroidism accounted for two-third of all the thyroid disorders observed among the patients. This is in sharp contrast to what was found 33 years ago in UCH, Ibadan where hyperthyroidism accounted for less than one-third of all the thyroid disorders [3]. The present study was not designed to examine the effect of iodine intake on the pattern of thyroid disorder in the population under review. However, it is possible that the universal salt iodization (USI) policy adopted by the Nigerian government might have a bearing to the relatively greater proportion of patients with hyperthyroidism in the present study compared to the study in UCH before the implementation of the USI programme [3]. This view is reinforced by the report of several recent studies which have conclusively shown that the risk of hyperthyroidism is increased in chronically iodine-deficient individuals who are exposed to sharp increases in iodine intake.
Considering the recent successful USI implementation policy in Nigeria, [15] this scenario was possible and may partly explain the increased incidence of hyperthyroidism. As in other reports [8, 14], the present study revealed that majority of thyroid disorders presented during the period of adolescence with 11.2 years as the mean age at presentation. In contrast, the study in UCH about three decades ago, reported a much lower mean age (5.4 years) at presentation [3]. This difference might be due to the fact that 60% of the cases in their series had congenital hypothyroidism and half of them presented before the age of six months with cretinism. The age range of their patients was 4 months to 14 years compared to 3.5 to 17 years in the present study. The absence of infants with cretinism in the present study might be related to the implementation of the USI policy by the Nigerian government [15]. The unique features of the patients seen in the present study were late presentation with florid signs of either hypothyroidism or hyperthyroidism and poor clinic attendance. Similar observation has been documented in a previous study [3]. The reason for the late presentation might be due to a general lack of awareness concerning endocrine disorders in our society; a factor that has been previously emphasized by Famiyuwa in UCH, Ibadan [16]. This view is supported by the comment made by the mother of the child with hypothyroidism (probably congenital) who presented at the age of 6 years. When this mother was asked why she delayed seeking medical help she said, “I did not notice it on time.” Also she had hoped the child will overcome those developmental challenges with time (the index patient is her only male child out of four children). The well known insidious onset of hypothyroidism may have contributed to the mother overlooking the presenting features of hypothyroidism in her only son until it became florid [1,2]. Our study revealed that duration of symptoms before presentation was shorter in hyperthyroidism compared to hypothyroidism, suggesting that patients with hyperthyroidism tended to present comparatively earlier than patients with hypothyroidism. This finding may be explained by the more insidious onset of hypothyroidism compared with hyperthyroidism. In addition, the presence of goiter might have contributed to the relatively shorter duration of symptoms before presentation in the case of hyperthyroidism compared to nongoitrous hypothyroidism. This in keeping with the social and cosmetic implication of the presence of goiter, particularly for teenage girls whose parents might be considering giving them out in marriage in the near future. The high clinic default rate made it difficult to document outcome of treatment and subsequent smooth transfer to adult physicians for continuation of care. This not surprising because Famuyiwa has emphasized that majority of their patients
(adults) were also lost to follow-up making it difficult to document outcome of treatment [17]. The principal clinical features observed among patients with hypothyroidism in our study included growth retardation, mental retardation, delayed language development and absence of goiter. These clinical features are in tandem with those reported among the patients seen in UCH, Ibadan [3]. There were no cases with thyroid nodules whether solitary or multiple among the patients seen in the present study. This observation is in agreement with the report from the study at the UCH, Ibadan, suggesting that thyroid nodules are not common in childhood and adolescence [3].

The most common thyroid disorder observed among the adolescents was hyperthyroidism. This is in contrast to other reports which stated that the most common presentation of thyroid disorders in adolescence was asymptomatic goiter [18]. The reason for this difference is not clear. Inadequate laboratory facility for detailed evaluation of endocrine disorders has been previously documented as one of the major management challenges with regard to practice of endocrinology in developing countries [16]. It would have been worthwhile to investigate for Peroxidase deficiency because this is one of causes of congenital hypothyroidism with insidious onset [2]. In the only case of euthyroid goiter, it would have been useful to establish the presence or absence of thyroid antibodies but we could not do this because of lack of laboratory facility.

CONCLUSION:

Hyperthyroidism constituted the highest proportion of the thyroid disorders seen between 2005 and 2011 and most patients with thyroid disorders tended to present late. High clinic default rate was a major management challenge.

REFERENCES:


KNOWLEDGE, ATTITUDES AND PRACTICES OF CAREGIVERS OF PATIENTS WITH SCHIZOPHRENIA IN PORT MORESBY, PAPUA NEW GUINEA

MALUO MAGARU

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ABSTRACT:

The purpose of this study was to investigate the knowledge, attitudes and practices of adult caregivers of schizophrenia patients in Port Moresby, Papua New Guinea. A semi-structured questionnaire was used to obtain the required data from consented caregivers accompanying the schizophrenia patients attending the consultation clinic at Port Moresby General Hospital (PMGH). Focus Group Discussion was held with consented caregivers of schizophrenia patients admitted in the Psychiatric ward of PMGH. A convenient sample of 79 caregivers with patients was selected for this study. Consents were obtained from 40 caregivers, which give a response rate of 50.6%. Most of the caregivers (75.0%) had no prior knowledge about schizophrenia; they became aware about schizophrenia from experience with their patients. The fathers (35.0%) and mothers (22.5%) were the major caregivers for the patients. Most of the caregivers indicated that the health workers did not give them adequate education about schizophrenia. Marijuana (47.5%) and psychosocial problems (45.0%) were the two major causes of schizophrenia indicated by the caregivers. Caregivers considered medical intervention to be the most important, but they also advocated supportive interventions such as church activities and family support. Most of the caregivers accepted their patients as part of the family and considered it their responsibility (52.5%) to care for them, and preferably to administer treatment at home (52.5%); the exception being during severely aggressive episodes. Financial problem was one of the factors that impacted negatively on follow-up of patients. Increased access to services and health education is recommended to further enhance the care of schizophrenia patients in Port Moresby, PNG.

Keywords: Schizophrenia, Caregivers, Knowledge, Attitudes, Practices, Papua New Guinea.

(Submitted December 2011, Accepted April 2012)
INTRODUCTION:

The schizophrenic disorders are characterized in general by fundamental and characteristic distortions of thinking and perception, and affects that are inappropriate or blunted [1]. Clear consciousness and intellectual capacity are usually maintained although certain cognitive deficit may evolve in the course of time. The most important psychopathological phenomena include though echo; thought insertion or withdrawal; thought broadcasting; delusional perception and delusions of control; influence or passivity; hallucinatory voices commenting or discussing the patient in the third person; thought disorders and negative symptoms [1]. There are sub types of schizophrenia; paranoid, hebephrenic, also called disorganized, catatonic, undifferentiated, simple and residual schizophrenia [1]. Paranoid schizophrenia is dominated by relatively stable, often paranoid delusions, usually accompanied by hallucinations, particularly of the auditory variety, and perpetual disturbance. Catatonic schizophrenia is dominated by prominent psychomotor disturbance that may alternate between extremes such as hyperkinesias and stupor, or automatic obedience and negativism [1]. Hebephrenic schizophrenia is a form of schizophrenia in which affective changes are prominent, delusions and hallucinations fleeting and fragmentary, behaviour irresponsible and unpredictable, and mannerisms common [1].

Undifferentiated schizophrenia is psychotic conditions meeting the general diagnostic criteria for schizophrenia but not conforming to any of the subtypes, or exhibiting the features of more than one of them without a clear predominance of a particular set of diagnostic characteristics [1].

Residual schizophrenia is a chronic stage in the development of a schizophrenia illness in which there has been clear progression from an early stage to the later stage characterized by long-term, though not necessarily irreversible, “negative” symptoms, e.g., psychomotor slowing; under-activity; blunting of affect; passivity and lack of initiative; poverty of quality or content of speech; poor nonverbal communication by facial expression, eye contact, voice modulation and posture; poor self-care and social performance [1]. Simple schizophrenia is a disorder in which there is an insidious but progressive development of oddities of conduct, inability to meet the demands of society, and decline in total performance [1]. The characteristic negative features of residual schizophrenia (e.g. blunting of affect and loss of volition) develop without being preceded by any overt psychotic symptoms [1].

According to the WHO, schizophrenia is a severe form of mental illness affecting about 7 per thousand people, mostly in the age group 15-35 years [2]. Schizophrenia affects about 24 million people worldwide and 90% of the untreated cases of schizophrenia are in the
developing world [2]. Schizophrenia is mainly characterized by chronic psychotic symptoms such as delusions, hallucinations and negative symptoms such as social withdrawal [3]. The aetiology of schizophrenia is multi-factorial, including genetics, structural brain abnormalities and neurotransmitter imbalance [3]. With prompt treatment, about a third of patients may recover fully from first episode and never have another episode, another third may improve with occasional relapses and others may follow a chronic, deteriorating course [3]. Without treatment the prognosis for most patients is poorer.

Schizophrenia, from the public health perspective, is a major concern as the onset of the illness occurs early (15-35 years of age) [2]. According to WHO reports it may affect about 1.0% of the general population in any given country [2]. This means that, in PNG with a population of 6.5 million, approximately 60 000 people may suffer from schizophrenia. It is a major public concern because it causes chronic disability, family disappointments, marital problems, financial disadvantages and destroys the education opportunities for those that are affected by the illness at early age. If the patient is a parent and the only bread winner for the family, then it creates an atmosphere of disability for the whole family. The education of the children is usually affected because the sick parents are emotionally detached from the family unit. It destroys the families’ hopes when the child they invest in is affected by the illness. The families may have more concern for the other children who are well and neglect the patient. Because of social stigma the communities may avoid the family members. The patients may be called names causing them to become violent or isolated in the community. There may be restrictions from community participation due to the disability that the illness causes. The impact of the stigma may prolong the duration of recovery of the patient. The illness may cause economic burden to the family as the patient may depend very much on financial support from close family members. The illness may create unemployment for the entire family. An employed patient may eventually lose his or her job because of stigma and low work performance. Hospital services usually have huge budgets to cater for the patients as the recovery periods are long.

Caregivers face problems of coping with the social withdrawal, awkward interpersonal behaviours and disruptive attitude of patients with schizophrenia. These behaviours may be unhealthy for the caregivers and may create a lot of stress and emotional discomfort in the families [2]. Caregivers are usually close family members that take the responsibility of looking after the sick patient in the family. They report on any changes or abnormalities that are observed with the patients during the follow-up visits to the health facilities [4]. The causes of the changes or abnormalities seen with the patients
may not be very clear to the caregivers but they may have their own interpretation of the abnormalities in their culture and they have some local names for the illness [4]. Caregivers may adapt to the environment of the schizophrenic patient and may live with them until there is a danger to the patient themselves or to the community; the caregivers have the responsibility of seeking help for the patient. In some cases schizophrenic patients are on the streets wandering aimlessly and surviving on what they find. This may be of concern to the caregivers who have the responsibility of caring for the patient; the schizophrenic patients are usually not considered responsible for their own actions.

Schizophrenia patients, when discharged, are managed at home by caregivers and are followed up at consultation clinics for maintenance of treatment and assessment. Early intervention and treatment are critical to prevent long term effects of the illness. The treatment is more effective when caregivers are equipped with the proper knowledge, attitude and practices relating to schizophrenia [5].

There have not been any studies on the knowledge, attitudes and practices (KAP) of caregivers of schizophrenia patients in PNG. Therefore, this study was done to assess the knowledge, attitudes and practices of caregivers of patients with schizophrenia, admitted in the PMGH in Port Moresby, PNG.

**PATIENTS AND METHODS:**

This was a cross-sectional descriptive study carried out in the psychiatric consultation clinic and the psychiatric ward (ward 6) in PMGH. The clinic is accessible to discharged psychiatric in-patients and new psychiatric outpatient referrals from health facilities in PNG. The study population was made up of caregivers accompanying schizophrenia patients to the psychiatric consultation clinic in PMGH. It was a convenient sample of 79 caregivers. The study was conducted between February and March 2010. Self designed semi-structured questionnaire was used to collect quantitative and qualitative data by interview during clinic sessions. Some of the information collected included: age, gender, marital status, employment status, level of education, religion, region of origin, relationship of caregiver to patient, and duration of the illness. Some of the data collected during the interview were written in the language that the caregivers preferred, in some cases the sessions were tape recorded. All data were thematically translated into English and analysed.

Ethical clearance and permission for this study were obtained from the ethics and research grant committee in the School of Medicine and Health Sciences (SMHS), University of Papua New Guinea (UPNG). Written consent was obtained from the Acting Chief Executive Officer of PMGH. Written consent was also obtained from caregivers before the interviews were conducted. Verbal consent was obtained from the guardians of schizophrenia.
participants before their participation in the focus group discussion.

All schizophrenic patients that were not accompanied by caregivers were excluded from the study. The quantitative data were analysed statistically using EPI-Info. 6.02. The qualitative data were analysed using Thematic Method. The main categories used in this study were, knowledge of the caregivers of the information of schizophrenia and its types, attitudes of caregivers towards the patients and the practices of caregivers.

RESULTS:
During the two months duration of this study a total of 565 appointments were booked, of which 328 were bookings for schizophrenia patients. Further analysis of the data indicated that there were 216 clinic attendances by 108 confirmed schizophrenia patients, because some of them attended more than once during the study period. A total of 29 schizophrenia patients were excluded from the study because they came alone to the clinic. The 79 caregivers and patients were enrolled for the study. Informed consent was obtained from 40 of the 79 caregivers, which gives a response rate of 50.6%. Gender distribution of the caregivers indicated 60.0% males and 40.0% females. Most of the caregivers (72.0%) were over 40 years of age. Information on marital status indicated that 80.0% of the caregivers were married, 10.0% were single, 7.0% widowed and 2.0% were divorced. For their residential status, 90.0% of the caregivers were home owners and 10.0% lived with their relatives. The caregivers were mostly from the southern region (75.0%), followed by the highlands region (15.0%), Niugini Islands (7.5%) and Momase (2.5%). Table 1 shows the relationship of the caregivers to the patients, the level of education and religion of the caregivers. The parents (57.5%) were the major caregivers, with the fathers (35.0%) playing the leading role compared to the mothers (22.5%). There was great variation in the educational level of the caregivers, with the majority having primary (27.5%) and tertiary/university (27.5%) education. Most of the caregivers were in the United Church (50.0%), followed by Catholic Church (22.5%). When caregivers were asked about their knowledge of schizophrenia and the source of their information, 75.0% had no prior knowledge about the illness and got the information first hand by patients experience; however 12.0% got the information from friends, 10.0% got it from health workers and 3.0% from the local news paper. When the caregivers were asked if the illness is only one type or different types, 90.0% indicated that there were many types of the illness, 7.5% indicated that there was one type and 2.5% were not sure. To assess their knowledge of recognizing schizophrenia, caregivers were asked to describe the different symptoms of their patients. Table 2 shows the different symptoms indicated by the caregivers. Physical
aggression (65.0%) and antisocial behaviour (65.0%) were the most frequent, followed by hearing voices, talking to self (60.0%), isolation and withdrawal (47.5%). Table 2 also shows the supportive interventions that caregivers recommended for the patients. Church (42.4%) and family support (40.0%) were highly recommended, followed by others such as, rehabilitation, stopping smoking, awareness and counselling. The caregivers were asked what they think caused the patient’s illness. Marijuana (47.5%) and psycho-social problems including divorce and others (45.0%) were the most frequently suggested causes; spirit (5.0%) and sorcery (2.5%) were the others suggested. When asked about the importance of medication for the patients, 92.5% of the caregivers indicated that medication was important. In response to question about awareness, 52.5% of the caregivers said there was enough awareness about schizophrenia, compared to 47.5% that said there was not much awareness in the community. Table 3 shows the response of caregivers on questions relating to their attitude to the patients. Most of the caregivers (27.5%) felt comfortable discussing the illness of the patient with others. The caregivers feel more comfortable talking to the family members (60.0%), communities (40.0%) and relatives (32.5%) when the patient relapses. Table 3 also show the attitude and reactions of caregivers when the patients make bizarre statements or bizarre actions. Being upset, calming and giving medication to the patient making bizarre statement or acting abnormally were highly expressed by the caregivers. When asked about witchcraft intervention, 75.0% of the caregivers do not believe in the witchcraft intervention, 7.5% said they have tried but failed and 5.0% said its long time intervention. Failed efforts included giving of herbs to the patients by the magicians. The caregivers were also asked about the preferred place for the patient to live; 52.5% said they would like the patient to stay in the house at home, 25.0% preferred the hospital and 22.5% preferred the village. Almost all people living in Port Moresby also have homes back in their traditional village. Therefore, some would keep the patient with them in their current home in Port Moresby. Others preferred to get them out of the city and take them back to the village. The question, “How you feel about caring for the patient?” was use to partly assess the attitude and responsibility of the caregivers towards caring for the patient. The responses of the caregivers are presented in Table 3. Most (52.5%) of the caregivers said that they were responsible to the patients. Figure 1 shows the responses of the caregivers when they were asked who do they see first when the patient relapses. The health worker was the first person seen by most (77.5%) of the caregivers, only 5.0% indicated that the police was seen first.
Table 1: Relationship to patients, educational level and religion of caregivers (n = 40)

<table>
<thead>
<tr>
<th>Relationship of caregivers with patients</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Father</td>
<td>35.0 (14)</td>
</tr>
<tr>
<td>Mother</td>
<td>22.5 (9)</td>
</tr>
<tr>
<td>Brother</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Relative</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Husband</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Sister</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Son</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Wife</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Daughter</td>
<td>2.5 (1)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Level of education of caregivers</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Tertiary / University</td>
<td>27.5 (11)</td>
</tr>
<tr>
<td>Primary School</td>
<td>27.5 (11)</td>
</tr>
<tr>
<td>High School</td>
<td>22.5 (9)</td>
</tr>
<tr>
<td>Elementary School</td>
<td>17.5 (7)</td>
</tr>
<tr>
<td>Secondary School</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>No education</td>
<td>2.5 (1)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Religion of caregivers</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>United Church</td>
<td>50.0 (20)</td>
</tr>
<tr>
<td>Catholic Church</td>
<td>22.5 (9)</td>
</tr>
<tr>
<td>Seven Days Adventist (SDA)</td>
<td>15.0 (6)</td>
</tr>
<tr>
<td>Pentecostal Church</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Describe the symptoms of the illness of the patients</td>
<td>Percent (n)</td>
</tr>
<tr>
<td>-----------------------------------------------------</td>
<td>------------</td>
</tr>
<tr>
<td>Abnormal belief</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Scared</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Sleep a lot</td>
<td>7.5 (3)</td>
</tr>
<tr>
<td>Verbal aggressive</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Abnormal speech</td>
<td>15.0 (6)</td>
</tr>
<tr>
<td>Smoking</td>
<td>15.0 (6)</td>
</tr>
<tr>
<td>Insomnia</td>
<td>15.0 (6)</td>
</tr>
<tr>
<td>Abnormality of mood</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Roaming/ wandering</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Self neglect</td>
<td>25.0 (10)</td>
</tr>
<tr>
<td>Isolation/withdrawn</td>
<td>47.5 (19)</td>
</tr>
<tr>
<td>Hearing voices/taking / laughing to self</td>
<td>60.0 (24)</td>
</tr>
<tr>
<td>Antisocial behavior</td>
<td>65.0 (26)</td>
</tr>
<tr>
<td>Physical aggression</td>
<td>65.0 (26)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>What supportive intervention is needed for the patient?</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Finance</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Give smoke</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>School</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Counseling</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Awareness</td>
<td>7.5 (3)</td>
</tr>
<tr>
<td>Stop smoking</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Rehabilitation</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Family support</td>
<td>40.0 (16)</td>
</tr>
<tr>
<td>Church</td>
<td>42.5 (17)</td>
</tr>
</tbody>
</table>

NB: All items are multiple response so do not add up to 100%
Table 3: Caregivers responses to attitude towards the patients (n = 40)

<table>
<thead>
<tr>
<th>Perception of Illness</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Discuss if asked</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Ashamed</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Relieve pressure</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>No discussion</td>
<td>17.5 (7)</td>
</tr>
<tr>
<td>Discuss for assistance</td>
<td>17.5 (7)</td>
</tr>
<tr>
<td>Awareness</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Comfortable to discuss</td>
<td>27.5 (11)</td>
</tr>
</tbody>
</table>

Which people do you feel comfortable talking to about the patient?

<table>
<thead>
<tr>
<th>People</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bosses</td>
<td>2.5 (10)</td>
</tr>
<tr>
<td>Health workers</td>
<td>15.0 (6)</td>
</tr>
<tr>
<td>Church members</td>
<td>27.5 (11)</td>
</tr>
<tr>
<td>Relatives</td>
<td>32.5 (13)</td>
</tr>
<tr>
<td>Communities</td>
<td>40.0 (16)</td>
</tr>
<tr>
<td>Family members</td>
<td>60.0 (24)</td>
</tr>
</tbody>
</table>

How do you react when the patient makes bizarre statements, comments or action?

<table>
<thead>
<tr>
<th>Reaction</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Give medicine</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Scared</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Ashamed</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Sad</td>
<td>30.0 (12)</td>
</tr>
<tr>
<td>Upset</td>
<td>30.0 (12)</td>
</tr>
<tr>
<td>Feel responsible calmly correct them</td>
<td>30.0 (12)</td>
</tr>
</tbody>
</table>

How do you feel about caring for the patient?

<table>
<thead>
<tr>
<th>Feelings</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Upset</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Repay what he has done</td>
<td>7.5 (3)</td>
</tr>
<tr>
<td>Happy</td>
<td>7.5 (3)</td>
</tr>
<tr>
<td>Burden</td>
<td>7.5 (3)</td>
</tr>
<tr>
<td>No problem</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Tiring</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Our responsibility</td>
<td>52.5 (21)</td>
</tr>
</tbody>
</table>

NB: Some items are multiple response so do not add up to 100%
Table 4: Willingness and barriers to attending consultation clinics; and home supervision of medication for patients (n = 40)

<table>
<thead>
<tr>
<th>How regularly are you able to attend the consultation clinic?</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Always</td>
<td>55.0 (22)</td>
</tr>
<tr>
<td>Most times</td>
<td>40.0 (16)</td>
</tr>
<tr>
<td>Some times</td>
<td>5.0 (2)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>What are some of the problems that stop you from coming to the consultation clinic?</th>
</tr>
</thead>
<tbody>
<tr>
<td>Forget dates</td>
</tr>
<tr>
<td>Other illness apart from schizophrenia</td>
</tr>
<tr>
<td>No escort</td>
</tr>
<tr>
<td>Distance</td>
</tr>
<tr>
<td>Non compliance</td>
</tr>
<tr>
<td>Aggressive</td>
</tr>
<tr>
<td>Surplus of antipsychotic</td>
</tr>
<tr>
<td>No problem</td>
</tr>
<tr>
<td>Patient refuses</td>
</tr>
<tr>
<td>Finance</td>
</tr>
<tr>
<td>Transport</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Who supervises the treatment at home?</th>
</tr>
</thead>
<tbody>
<tr>
<td>Respondent caregivers</td>
</tr>
<tr>
<td>Parents of patients</td>
</tr>
<tr>
<td>Patients themselves</td>
</tr>
<tr>
<td>Siblings of patients</td>
</tr>
</tbody>
</table>

NB: Some items are multiple responses so do not add up to 100%

Fig. 1: Who do you see first when the patient relapses? (n = 40)
Table 5: Difficulties for caregivers and supportive interventions for patients recovery (n = 40)

<table>
<thead>
<tr>
<th>Difficulty</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wander/ roaming streets</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Very old</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Demand food</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>Doesn’t listen</td>
<td>5.0 (2)</td>
</tr>
<tr>
<td>No problem</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Non compliance</td>
<td>17.5 (7)</td>
</tr>
<tr>
<td>Money</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Demand smoke</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Aggressive/violent</td>
<td>47.5 (19)</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Activity</th>
<th>Percent (n)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Early detection of break down</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Clean environment</td>
<td>2.5 (1)</td>
</tr>
<tr>
<td>Awareness</td>
<td>10.0 (4)</td>
</tr>
<tr>
<td>Stop smoking</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Counselling</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Finance</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Church involvements</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>Employment</td>
<td>12.5 (5)</td>
</tr>
<tr>
<td>School</td>
<td>17.5 (7)</td>
</tr>
<tr>
<td>Rehabilitation</td>
<td>20.0 (8)</td>
</tr>
<tr>
<td>Family support</td>
<td>25.0 (10)</td>
</tr>
<tr>
<td>Medicine</td>
<td>37.5 (15)</td>
</tr>
</tbody>
</table>

NB: Some items are multiple responses so do not add up to 100%

The responses of caregivers to questions related to willingness and problems or barriers to attending consultation clinics are presented in Table 4. Fifty five percent of caregivers said they attended the clinic always, compared to 40.0% who attended most times and 5.0% that only attended sometimes. The major problems that prevented some caregivers from bringing their patients to the consultation Clinics, included finance (27.5%), transport (27.5%) and refusal by the patients (27.5%). Having surplus of antipsychotic drugs at home (7.5%) and aggressive behaviour of patients (7.5%) were other problems mentioned. Responses to
supervision of medications for patient at home are also presented in Table 4. The caregivers (57.5%) were mostly responsible for the supervision of medication at home. The patients themselves supervised their own treatment in 12.5% of cases. However in some cases, supervising medication was not the responsible of the respondent who had brought the patient to the consultation clinic, but was the responsibility of other family members such as parents of patients (25.0%) and siblings (5.0%) of patients. Table 5 shows the responses of caregivers to questions about the difficulties they faced and suggested activities to support recovery of the patients. Aggression and violence (47.5%) were the most common difficulties faced by caregivers. Demand for cigarettes (20.0%) and money (20.0%) and also non compliance with medication (17.5%) were other difficulties mentioned. Medication (37.5%), family support (25.0%), rehabilitation (20.0%) and school (17.5%) were seen to be the most support activities for recovery.

Results of the focus group discussion (FGD):
FGD was conducted in ward six at PMGH. There were eleven inpatients during the time of FGD, but only five were schizophrenia patients. Prior to the FGD, the five guardians were approached and all agreed to participate. All the respondents were males, the mean age was 29.6 years and the age range was 20 to 50 years. Each of the five respondents had different occupation: farmer, principle, pastor, apprentice and student. Each respondent was closely related to the patient, father, uncle, aunty and brother. During the FGD the participants were asked the same questions as caregivers at the consultation clinic but in a group setting. The answers in general were similar to those given by the caregivers; the same points were raised and generally agreed upon by the participants in the FGD.

DISCUSSION:
The interviews and FGD were conducted by a trained mental health professional who is a Health Extension Officer that is conversant with the cultural settings and local languages in the study area. This minimises the negative impact of translation and interpretation of expressions used by the caregivers and participants in the FGD. It was also easy to understand slangs and colloquial terms used by the respondents. Results obtained in the present study are similar to the findings reported in other studies conducted in India [6], Cambodia [7], Asia [8], Nigeria [9] and Australia [10].

Our data indicate that schizophrenia patients accessed the consultation clinic more often than patients suffering from other mental illnesses. Caregivers' knowledge on schizophrenia illness indicates that they understand the general idea of the illness as mental sickness. The term schizophrenia to them was ‘longlong’ (mental illness). Eighty percent of caregivers said that, they had heard...
of schizophrenia, however the real knowledge on the impact of the illness cannot be significant as the caregivers described the illness as many forms.

Most respondents said that they knew of the illness from the patient’s experiences, some from the health workers and others through friends. The term schizophrenia was new to them but the understanding of the illness from the caregivers’ perspective was not restricted to the term schizophrenia but to general mental illness. The caregivers said that living with the patient taught them many things about the mental illness.

The respondents said that there were many types of schizophrenia. Three of the caregivers were not sure of the diagnosis while one said there was only one type. This finding illustrates that caregiver’s lack accurate knowledge of schizophrenia.

Descriptive classification is one of the systems mostly used in the case of diagnosing of schizophrenia and other mental disorders [11]. Most caregivers in this study can identify the acute stage and the prolific stages. They have described the negative and positive symptoms of schizophrenia well with all symptoms that are seen in the patients. They agreed that there are many types of schizophrenia with only three out of forty respondents being unsure of the illness. All respondents described the different forms of schizophrenia in many ways. Physical aggression together with anti social behaviour, hearing voices, taking to self, laughing to self and isolation were regularly mentioned in descriptions of schizophrenia. Self neglect, roaming the streets, abnormality of mood, sleepless nights (insomnia), smoking, abnormal speech and verbal aggression were also stated. The other least described features were sleeping a lot, feeling scared and abnormal belief. The respondents were of the view that medication was the only necessary intervention for this illness. Apart from medicine being the major intervention, the other supportive interventions that can support the patients were church activities, family support, rehabilitation, and stopping smoking, counselling and going back to school. Others responded that giving of smoke and money to the patients was also a helpful intervention.

Fifty two percent of caregivers said that health workers were doing enough education on schizophrenia during consultation while 48% said that not much education on schizophrenia was done during consultation. The response to the education on schizophrenia awareness by the caregivers did not differ very much as nearly half (48%) felt health workers were not giving them enough education. The similar findings were reported in the National alliance of Mental Illnesses in 2008.

Most respondents said they felt very comfortable to discuss the patient’s illness with others. The reasons for discussing patients’ issues were for the reasons of awareness and assistance. Others felt that discussing the
patients’ illness with others relieved them from the problems that they keep within themselves. Others didn’t want to discuss with others unless they were asked. Others were ashamed to discuss the patients’ illness. Most of the respondents felt very comfortable talking about the patients’ illness to close family members, communities, relatives, church members and health workers. The importance of this is that caregivers with less stress help the patient in whatever way they can, more than the stressed caregiver who is under pressure and does not help the patient. The patient whose caregiver is under pressure is most likely to be neglected and the patient is most likely to relapse [12].

Behavioural problems in patients are the most common difficulties that caregiver encounter. The most difficult that they face is aggression, anti social behaviours like demanding cigarettes, money, food, wandering on the streets and the patient not obeying the caregivers.

Other problems noted were that caregivers felt they were too old to care for the patient. The greatest difficulty that is faced by the caregivers as mentioned is aggressiveness. Frequently violent behaviour is aggravated by the caregivers as they try to control the patients. The patients demand much from the caregivers which the caregivers don’t have, so they have to explain the reasons for not providing what the patients need. Disagreement causes irritability of the patients. The difficulties faced by the caregivers to sustain the demands for the patients are real. We depend on whatever we sell to sustain ourselves and the family or receive donations from neighbours. The first people to be consulted when the patient relapses are health workers. The respondents also said that close relatives, police and priests or pastors were consulted when the patients relapsed. Caregivers living in the city are aware of the health care that is provided for the patients. Unfortunately easy access to psychiatric hospitals is only possible for a minority of the population as most of the population live in rural areas.

The reasons for not bringing the patients to the consultation clinic were transport, finance and patients refusing to come to the clinic. Patient refusing to come to the clinic means lot of things such as non compliance, long waiting time at the clinic or maybe due to staff approaches to the patients. The other constraints were surplus of medicine, distance, no escorts, forgetting of the dates and being sick with other physical illnesses. With or without the above mentioned problems, the respondents said that they are happy to attend to the consultation clinic.

Most caregivers said that they were responsible for supervision of the treatment at home. Other said that patients used to supervise their own treatment. The respondents said that they preferred to supervise treatment. Maintenance of treatment and rehabilitation at home is
important and should be encouraged. Caregivers should be made aware of the early signs of relapse so that when a patient shows the signs he or she should be brought to the clinic or nearby health facility for the patient to be referred.

CONCLUSIONS:
This study set out to investigate knowledge, attitudes and practices of caregivers of schizophrenia patients. Most caregivers had heard about schizophrenia; patient experience was the main source of information in understanding schizophrenia; marijuana and psychosocial factors were identified as the main causes of schizophrenia; medication was the most important intervention and not intervention such as witchcraft; supportive interventions included church activities, family support, rehabilitation, school, employment and finance; not much awareness has been done on schizophrenia illness in communities; it was comfortable to discuss their relative’s illness with others for the sake of awareness and assistance; it is important to share experiences with family members, communities, church members and health workers; it is necessary to take responsibility for the patient and supervise their treatment at home, with the exception of patients that are aggressive and could not be handled at home;

Financial limitations, transport, patient’s refusal, surplus of antipsychotic medication, noncompliance were the main factors for non attendance to the consultation clinics; Responsible caregivers should not be denied the truth that their family member is suffering from schizophrenia; health workers should be encouraged to share their knowledge, skill and experience with communities.

The caregivers are willing to take the responsibility of looking after their loved ones but the responsibility of imparting the knowledge, shaping the attitude and practice relies heavily on the service providers.

ACKNOWLEDGEMENT
The Human Resource Branch of the National Department of Health in Papua New Guinea is acknowledged for their sponsorship and the management of Laloki Psychiatric Hospital for proving the research grant for this study. Many thanks to Dr. Florence Muga, Professor Paul Crouch-Chivers, Dr. Roger Nascimento, Mr. Russell Kitau, Dr. Simon Mete, Dr. Ludwig Nanawar, Dr. Monica Hagali, Dr. Goiba Tienang, Dr. Ngason, Dr. Maukera, Dr. Quintao, Mr. Lama Rema, Sr. Doris Pairua, Sr. Hirata Dobadoba, CHW Jessie Milli for their encouragement and support. Thanks also to Mr. Lope, Mr. Ase, Mr. Vetari, Mr. Symakus, Sr. Muru and the ward six staff of PMGH. Very special thank you to the families and patients who participated in this project;
REFERENCES:


EFFICIENCY OF SODIUM HYPOCHLORITE AND FOUR OTHER INTRA CANAL MEDICAMENTS IN ELIMINATING THE CANDIDA ALBICANS IN THE ROOT CANAL SYSTEM – AN EX-VIVO EVALUATION

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ABSTRACT:
Evaluation of the effect of 2.5% Sodium Hypochlorite and four other intra-canal medications on Candida albicans harvested from root canals. The contaminated canals were irrigated with sterile saline and then treated with either, Calcium Hydroxide + Saline; Calcium Hydroxide + 2.0% Chlorhexidine Gluconate; Zinc Oxide + 2.0% Chlorhexidine Gluconate; Amphotericin B powder + Distilled water, or 2.5% Sodium Hypochlorite. In one group (control) the root canals were not treated with any medication. Canal access and apex were sealed with Cavit and the roots were stored in an incubator at 36 to 38° C for 14 days. The canals were re-instrumented and irrigated with saline. Sterile paper points were used to transfer the root canal contents to test tubes containing saline. Part of the suspension was harvested on Sabouraud dextrose agar with Chloramphenicol and incubated at 36 to 38° C for 48hrs. Irrigation with 2.5% Sodium Hypochlorite was effective in 90% of the samples, Calcium Hydroxide + Saline was the least effective (50% effective). Within the limitations of this study, long term intra-canal medication was important to eliminate some Microorganisms especially C. albicans present inside root canal.

Key words: Amphotericin B, Candida albicans, Calcium Hydroxide, Chlorhexidine Gluconate, Sodium Hypochlorite.

(Submitted March 2012, Accepted May 2012)
INTRODUCTION:
The control and elimination of microorganisms are very important during endodontic treatment because of their role in pulpal and periodontal diseases. Presence of black-pigmented bacteroides in teeth with pulp necrosis, putrid smell and pain, were typical clinical symptoms associated with these microorganisms. Other symptoms like formation of fistula, swelling, and sensitivity to palpation were also related to these microorganisms [1]. The chemomechanical cleaning of root canal is one of the most important part of infection control process and can eliminate many of the microorganisms present, although some may remain in the root canal system since they have the ability to penetrate deeper into dentinal tubules, thus an intracanal medicament is necessary to eliminate them [2,3]. For selection of the proper intracanal medicament it is necessary to determine which microorganisms are present to destroy them as well as neutralize their byproducts. Calcium hydroxide alone or in combination with other medicament has been widely used in teeth with radiographic evidence of periapical lesions [4]. However there are some cases where conventional therapy does not eliminate these microorganisms from the root canal system [5]. Microbiological investigations of apical periodontitis have revealed that yeasts can be isolated from the root canal together with bacteria [6-8]. Using light microscopy nine cases of apical periodontitis resistant to endodontic treatment were analyzed. Six of the biopsies revealed microorganisms in the apical third of the root canal; four biopsies contained more than one bacteria species and two contained yeast [6]. A total of 48 fungal species were isolated from root canals of teeth with apical periodontitis [9]. The most common isolated species is Candida albicans since it has the ability to colonize deeper into dentinal tubule [10]. Other isolated species are C. glabrata, C. guilliermondii, C. inconspicua, and Geotrichum candidum. Yeasts can be detected in 7-18% of infected root canals and 7% of persistent root canal infections [11]. Since yeast has been isolated in teeth with pulpal infections we evaluated ex- vivo, the effect of 2.5% Sodium Hypochlorite and four other intracanal medications on C. albicans harvested inside root canals.

MATERIALS AND METHODS:
Sixty human freshly extracted single rooted teeth (non carious - closed apices) were used in this study. The crowns were sectioned and the average root size was about 16.0mm. The root canals were instrumented 0.5mm short of apex up to #25file. The roots were coated externally with 2 coats of nail polish, except the
cervical access and apical foramen, and autoclaved at 123°C for 30 minutes.

The remaining procedure was performed inside a laminar flux chamber using sterile materials and instruments. After this period the apical and cervical sealings were removed and the root canals were instrumented again up to #40 file using 5.0 ml sterile saline for irrigation. Root apices were sealed with Cavit cement and root canals were contaminated with 0.1 ml of a suspension containing \(10^8\) C. albicans cells using sterile micropipette. The access openings were sealed with sterile cotton pellet and Cavit cement and the roots were placed in a sterile receptacle and maintained in an incubator at 37±1°C for 48 hrs. The roots were then divided into groups of 10 each according to medicaments as shown in Table 1.

<table>
<thead>
<tr>
<th>Groups</th>
<th>Medicaments</th>
</tr>
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<tbody>
<tr>
<td>Group 1:</td>
<td>Ca(OH)(_2) powder + Saline</td>
</tr>
<tr>
<td>Group 2:</td>
<td>Ca(OH)(_2) powder + 2.0% Chlorhexidine Gluconate (CHX)</td>
</tr>
<tr>
<td>Group 3:</td>
<td>ZnO powder + 2.0% Chlorhexidine Gluconate (CHX)</td>
</tr>
<tr>
<td>Group 4:</td>
<td>Amphotericin B powder + distilled water</td>
</tr>
<tr>
<td>Group 5:</td>
<td>Irrigation with 2.5% NaOCl during the procedure with no intracanal medication</td>
</tr>
<tr>
<td>Group 6:</td>
<td>Roots are sealed with cavit with no intracanal medication (Control Group)</td>
</tr>
</tbody>
</table>

The roots were then incubated at 37±1°C for 14 days in an incubator. After 14 days, canals were again instrumented with #25 file and irrigated with saline to remove medicaments. Pure culture, isolated from the canals, were identified and confirmed by gram staining and germ tube testing. For microbiological evaluation sterile paper points (#25) were placed in the root canal for one minute, and then placed in a test tube containing 1.0 ml sterile saline, shaken for 30 seconds and 0.1 ml aliquots were harvested on Sabouraud dextrose agar plates with Chloramphenicol (1.0 mg/ml) and incubated at 37°C for 48 hrs. The SPSS was used for analyses of the data. Fisher test was used for statistical analysis and p-value < 0.05 was considered significant.
Figure A: Sterile paper points placed in a test tube containing 1.0ml sterile saline.

Figure B: Fungi growth on Sabourand dextrose agar plates with Chloramphenicol (1.0mg/ml) after 48 hours.
RESULTS:
The results obtained are presented in Table 2. The 2.5% NaOCl was effective in 90% of the samples, followed in decreasing order of effectiveness by Amphotericin B powder and distilled water (80% effectiveness), ZnO powder and 2% CHX (70% effectiveness), Ca(OH)$_2$ powder and 2% CHX (60% effectiveness), Ca(OH)$_2$ powder and saline (50% effectiveness), and saline + no intracanal medication.

Table 2: Positive and Negative growth of Candida albicans in the six groups

<table>
<thead>
<tr>
<th>Group 1</th>
<th>Group 2</th>
<th>Group 3</th>
<th>Group 4</th>
<th>Group 5</th>
<th>Control</th>
</tr>
</thead>
<tbody>
<tr>
<td>Positive</td>
<td>5 (50%)</td>
<td>4 (40%)</td>
<td>3 (30%)</td>
<td>2 (20%)</td>
<td>1 (10%)</td>
</tr>
<tr>
<td>Negative</td>
<td>5 (50%)</td>
<td>6 (60%)</td>
<td>7 (70%)</td>
<td>8 (80%)</td>
<td>9 (90%)</td>
</tr>
</tbody>
</table>

DISCUSSION:
The objective of endodontic therapy is to achieve a root canal free of microorganisms. This can be achieved by shaping and cleaning of root canal with the use of antimicrobial irrigants and intracanal dressings [12]. Sodium hypochlorite [13] has been widely used for irrigation. However some microorganisms may be resistant to conventional endodontic treatment. In the present study the 2.5% NaOCl used as an irrigant during instrumentation-showed best antifungal efficacy. The 2.5% NaOCl was used because it is known to posses similar antifungal effects but has low toxicity compared to the higher amount of 5.25% [14]. A total of 90% of samples showed negative C. albicans culture after treatment with 2.5% NaOCl solution. The growth of yeast in 10% of the samples may be due to the difficulty of penetration of this irrigant into root canal irregularities [15]. The root canals irrigated with 2.5% NaOCl, were not treated with an ICM, and also since EDTA was not used during instrumentation deeper smear layer may not have been removed, which otherwise might have enhanced the action of the chemomechanical preparation, thus eliminating all the microorganisms.

The control group, irrigated only with sterile saline showed C. albicans growth in 90% of the samples. The elimination of C. albicans in one control sample (10%) may be due to the topical effect of irrigation [16]. Second best result was shown by Amphotericin B, – (a commonly used
Amphotericin B is fungistatic as well as fungicidal depending on the concentration used. It acts by binding to the steroids in the cell membrane of fungi, with resultant changes in membrane permeability allowing leakage of intracellular components. Combination of ZnO and 2.0% CHX was more effective compared to Ca(OH)₂ and 2.0% CHX [18]. Ca(OH)₂ mixed with saline was least effective in eliminating C. albicans [19]. C. albicans has been demonstrated to be highly resistant to Ca(OH)₂ if used along with inert material such as Glycerine and saline [17]. Recent studies have suggested that CHX combined with Ca(OH)₂ is effective against Ca(OH)₂ resistant microbes [20]. Chlorhexidine is more effective than Ca(OH)₂ in eliminating C.albicans [21]. Since CHX is known to have substantivity, it showed slightly better result when mixed with Ca(OH)₂ compared to saline.

CONCLUSION:
This study reinforces the importance of endodontic treatment in two sessions with the use of long term intracanal medication to eliminate microorganisms present inside root canal.

C. albicans is an opportunistic microorganism and a poorly functioning immune system might increase the risk of fungal infection in the root canal space. This study also highlights the presence of C. albicans, which may persist even after endodontic treatment. Further evaluation may be necessary to find an irrigant along with a medicament which could eliminate all microorganisms under clinical situation.

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CASE REPORTS

PROGRESSIVE HEMIFACIAL ATROPHY- A CASE REPORT
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Running title: Progressive Hemifacial Atrophy

ABSTRACT:
Progressive Hemifacial Atrophy, also known as Parry-Romberg Syndrome, is an uncommon degenerative and poorly understood condition. It is characterized by a slow and progressive atrophy affecting mostly one side of the face along with ophthalmic and neurological complications. The disease is said to overlap with linear scleroderma. No dearth of speculation has been unturned to find the causative agent since it was first described. A case report of 35 years old, female is presented.

Key words: Progressive, Hemifacial, Atrophy, Parry-Romberg, Romberg,

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INTRODUCTION:
Among the diseases typically presenting unilaterally in orofacial region, one of the rare diseases is progressive hemifacial atrophy, also known as Parry-Romberg Syndrome (PRS). It is characterized by a progressive atrophy of the skin and subcutaneous tissue on one side of the face [1]. This rare condition was first described by Calef Parry in 1825 [2], later by Moritz Romberg in 1846 [3]. Romberg named the process “Trophoneurosis of the face,” emphasizing the implication of the nervous system in this syndrome [4]. The worldwide prevalence is unknown [5]; though Stone suggested one per half a million births [6]. The syndrome is more common in females, with onset in the first or second decade followed by a ‘burning out’ of the atrophic process and subsequent stability [7]. There is a predilection for the left side of the face [7]. Many causes have been proposed such as facial or body trauma, compression of the
cervical sympathetic nervous system, infection including Borrelia burgdorferi, autoimmune neurovascular disorders [7-9].

PRS is mostly diagnosed based on clinical signs and symptoms, characterized by hemifacial atrophy of the face, coup de sabre on forehead and with many oral dental manifestations such as atrophy of tongue, root resorption, tooth dilacerations [9]. Skin changes of PRS often resemble “en coup de sabre” of linear scleroderma [10].

This case study describes the clinical features of Parry Romberg Syndrome in a middle aged female. Written informed consent was obtained from the patient and ethical clearance and permission was obtained from the institutional ethical committee for the study.

CASE REPORT:
A 35 year old female patient reported to the Department of Oral Medicine and Radiology, with the complaint of gradual stiffness and hollowing of the right side of face since 6-7 years. There was no relevant medical and family history. On examination facial asymmetry was detected due to the hollowing of cheek and chin on the right side. The right eye was mildly depressed in the socket, and atrophy of facial skin and lower lip on the right side was observed. Depression of nasal bone on the right side was noticed. Because of the loss of subcutaneous fat there was prominent bony ridge on the ipsilateral side, when compared to the normal side. Hyperpigmentation was noticed on forehead and the chin region. Localised vitiligo patches were seen in the right ear. There was atrophy of temporalis, buccinator, masseter muscles and prominent zygomatic arch on the right side (Fig 1A). Demarcation between the affected and unaffected side was evident in the lower one third of the face (Fig 1B).

On palpation muscles were very firm, nontender. The intraoral examination revealed the atrophy of tongue on the right side.

All teeth appeared normal. Based on the clinical features, a diagnosis of PRS was made. Routine blood investigations were carried out which revealed all values within normal limits. Orthopantomogram and posterior-anterior cephalogram was done (Figs 2 & 3).
Fig. 1A: Extra oral view showing atrophy of muscles, vitiligo patches on the right ear. Fig. 1B: Wrinkled appearance of the overlying skin due to lack of subcutaneous fat on the right side.

Fig 2: Orthopantomogram shows no abnormality.

Fig. 3: PA cephalogram shows asymmetry of the jaws with severe atrophy of soft tissues on right side.
Except for small maxillary sinus on right side no abnormality was observed in orthopantomogram. The posterior-anterior cephalograms revealed asymmetry of the jaws with severe atrophy of soft tissues on right side. Alloplastic implants or omentum graft suggested to the patient for correction of aesthetic deformity, but the patient could not afford it for financial reasons. Psychological counseling was done for the patient as well as her relatives. They were assured of the benignness of the condition and explained about the course of the disease. The patient was followed up for a period of six months and no obvious changes were noticed.

**DISCUSSION:**

Hemi-facial atrophy is a rare disorder characterized by slowly progressive self-limited degeneration of the soft tissues of one half of the face (hemifacial atrophy) including dermis, subcutaneous tissue, fat, cartilage, and sometimes bone [11]. This condition is rarely manifested bilaterally. Along with the aesthetic abnormality there is functional disturbance often involving the eyes, neurological abnormalities. The disease may be associated to neurological abnormalities such as epilepsy, facial pain and migraine. Half of patients have eye or vision problems. Some develop autoimmune disorders like vitiligo and thyroid disease [5].

Symptoms and physical findings associated with PRS usually become apparent during the first decade or during the early second decade of life [12]. In some cases, the disorder is apparent at birth. Though in this case the patient noticed the abnormality in her third decade of life and in contradiction to most of the cases, the right side was affected. The degree of facial deformity is usually more severe if atrophy begins in the first decade, as growth is rapid during this time [9]. In the present case facial disfigurement has forced the patient to seek dental consultation rather than any functional disturbance. Clinically, the skin can be dry and hyperpigmented which is also seen in presented case. Some patients present with a demarcation line between normal and abnormal skin, known as “coup de sabre” [13], which was not seen in our case. Atrophy of underlying muscles, bones, and cartilage is responsible for the typically aged appearance of the patient. Though absent in the present case, blanching of the hair or bald patches on the scalp and loss of eyelashes and eyebrows may occur [14].

The skin overlying affected areas may become hyperpigmented with patches of vitiligo. Localised vitiligo patches were seen in the right ear. The ear can be misshapen, smaller than normal, or bat-eared as a result of tissue atrophy, none of these features were seen in the present case [10]. Anterior to posterior growth can be altered by deviation of the entire
middle and lower third of the face to the affected side, carrying the nose and chin with it. When the lips are involved, they can be showing a unilateral dentition. There may be atrophy of the upper lip and tongue, which was observed in our case [10]. Intraoral soft tissues and muscles of mastication can be affected, but they usually function normally. A decreased depth and width of the retromolar region of the pharynx may occur. Delayed eruption, missing teeth, deficient root development, or resorption of the roots of teeth, dilacerations of the tooth of affected side, have been reported [9]. The mandible and alveolar ridge may be smaller on the affected side. The mandibular body may be shorter than normal, the ramus can be deficient vertically, and there can be a delay of mandibular angle development. The jaw disturbances can result in a unilateral malocclusion on the affected side and deviation of the facial and dental midlines. Spontaneous fracture of the mandible has also been reported [9]. Rarely there may be associated involuntary jaw closure [15]. In the current case no dental abnormality was observed in the patient.

Some individuals may experience other associated symptoms like neurological abnormalities such as epilepsy, migraine and facial pain, trigeminal neuralgias, facial paresthesias, abnormalities of hair like alopecia, ophthalmic disorders such as heterochromia, uveitis, enophthalmos due to atrophy of fat around the eye [6]. Enophthalmosis was evident in the present case.

There is an age-old argument about the relationship between linear scleroderma andPRS [16]. Controversy remains regarding the relationship of PRS to linear scleroderma, mainly about considering them as distinct entity or including PRS in the spectrum of linear scleroderma [5]. In linear scleroderma, the lesions usually are limited to the skin and to the subcutaneous tissue beneath the cutaneous lesions; rarely, however, the underlying muscles and bones are also affected. In PRS, the atrophy is deeper than that seen in linear scleroderma. The skin is less often bound down. More extensive involvement of the lower face is another feature of PRS [17]. Skin biopsy of PRS is indistinguishable from that of linear scleroderma [16]. The histopathological characteristics of linear scleroderma consist of two stages including, early inflammatory and late sclerosis stages. There are no inflammatory changes in PRS, even in its early stages. The epidermis is normal; the collagen bundles in the reticular dermis often appear thickened and closely packed and stain more deeply eosinophilic than in normal skin; the eccrine glands appear markedly atrophied [18, 19]. There is no clear cut difference in autoantibodies between localized scleroderma and Parry Romberg syndrome. The coexistence of auto-antibodies such as anti-dsDNA in both the disorders may confirm that Parry Romberg syndrome and
linear scleroderma en coup de sabre represent overlapping conditions [16].

Management of hemifacial atrophy is multidisciplinary, including reconstructive surgeries, orthodontic treatment, and psychosocial support, management of ophthalmic and neurologic complications [20].

Reconstructive facial surgery employing techniques including lipo-injection, dermis fat grafting, silicone implants, tissue transfer are needed to repair wasted tissue and diminish facial asymmetry [6, 13] however the outcome is unpredictable.

Orthodontic treatment (corrective osteotomy) can help in the correction of any associated mandibular malformation [21]. None of these surgical treatments could be carried out in our patient because of financial reasons. Along with the surgical correction it is of paramount importance that these patients are provided with psychological support as it is very hard to cope with the gross facial deformity [9].

CONCLUSIONS:
PRS is an uncommon condition, which manifest as atrophy of one side of the face. The pathophysiology of the syndrome remains unknown. This case reports highlights the features of progressive hemifacial atrophy.

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The authors report no conflicts of interest related to this study.

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COCKAYNE’S SYNDROME: A CASE REPORT

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Running Title: Raghavendra et al. Cockayne’s syndrome

ABSTRACT:

Cockayne's syndrome (CS) is a rare, autosomal recessive disease resembling progeria. The features of CS do not appear until 4 to 5 years of age. Most patient presents with cachectic dwarfism, cutaneous photosensitivity, loss of adipose tissue, mental retardation, skeletal and neurological abnormalities, similar to the current case. The additional feature observed in the present case was actinic chelitis. We report a case of Cockayne’s syndrome with pronounced oral manifestations and an unusual feature of actinic chelitis.

Key words: Cockayne Syndrome, Actinic Cheilitis, Genetic Disorder, Progeria

(Submitted April 2012; Accepted June 2012)

INTRODUCTION:

Cockayne’s syndrome (CS) is a rare autosomal recessive disorder described first by Cockayne in 1936 [1]. It has been proposed that mutation of two genes, CNK1 (ERCC8) and ERCC6, located on the 5 and 10 chromosomes respectively may lead to two variations of CS [1]. The major features of CS include progressive loss of muscle and subcutaneous tissue, short stature, premature aging, senile face, mental retardation, microcephaly, retinopathy, beak-like nose, hearing loss, carious teeth, relatively large hands and feet, joint contractures, photosensitive dry skin, and thin hair [2]. Progression of CS leads to functional debility and bed ridden in the second decade of life. Patient eventually dies because
of respiratory and other infections [3]. Patients affected by this syndrome usually have unfavorable prognosis. Early diagnosis is most crucial, for proper genetic counseling and antenatal screening [4]. The present case is relevant because it is a rare condition and presents important oral findings.

CASE REPORT:

A 14 year old male patient, born of a consanguinous marriage, was brought to the hospital by his father for complaint of wound on the lower lip since two years. The wound was associated with provoked bleeding and moderate pain. Medical history revealed that, before visiting our department the patient was hospitalized for 10 days, as he was suffering from tremors and difficulty in speech. Patient’s father reported that the child was given treatment in the local hospital for chronic suppurative otitis media at the age of 5 years and 10 years. Neonatal history revealed lower segment cesarean term, birth weight 2.75kg and no post natal complications. Anthropometry revealed weight 15.5kg and height 119.0cm. Ataxia, cerebellar signs, gait abnormality, horse riding stance were present (Fig. 1). Magnetic resonance imaging of the head and brain showed mild cerebrum and cerebellum atrophy (Fig. 2); no abnormality was detected with chest and hand radiographs. Blood cell count and serological values were within normal limits. Ophthalmic evaluation revealed evidence of exposure keratitis started lacryge. On examination, an excoriated lesion with vertical fissuring and crustations were apparent on vermillion zone of lower lip and labial mucosa (Fig. 3). Gingiva was soft and edematous, partial macrodontia principally of central incisors, generalized dental fluorosis with hypoplasia and deep palate were noticed. Panoramic revealed bilateral condylar hypoplasia. Cytogenic reference was opined as cockayne syndrome. Actinic cheilitis was set as the provisional diagnosis for the lip lesion and patient was advised to apply sunscreen cream with sun protection factor 15. After a week the patient was reexamined, there was marked regression of the lesion. Patient was advised to avoid unnecessary exposure to the sunlight and to go for regular health check up. Patient informed consent and Ethical committee clearance was obtained for the present case report.
Fig 1: Height, gait and horse riding stance

Fig 2: MRI images of brain showing mild cerebrum and cerebellum atrophy

Fig 3: Photograph showing excoriated lesion in Vermillion zone of the lower lip with vertical fissuring and crustations.
DISCUSSION:
Xeroderma pigmentosum (XP), Cockayne syndrome (CS) and trichothiodystrophy (TTD) are genetic disorders with very different clinical features, but they are associated with defects in nucleotide excision repair [5]. For affected individuals at age of 2 to 4 years, CS usually becomes evident with changes in personality and behavior [4]. Mental deterioration is progressive and the dwarfism becomes obvious at this time. A characteristic facies develops, resulting in a thin prominent nose, prognathism, sunken eyes, and a lack of subcutaneous fat. Other major neurological abnormalities include sensorineural hearing loss, ataxia, spasticity, myoclonus, and gait disturbance [6]; similar characteristic findings were noticed in the present case.

We noticed macrodontia of central incisors, generalized dental hypoplasia and condylar hypoplasia in the present case. The usual oral findings are delayed deciduous teeth eruption, oligodontia, short roots, more incidence of caries, a deep palate, atrophy of the alveolar processes, mandibular prognathism and condylar hypoplasia [7].

The diagnosis was basically clinical, though supportive diagnostic tests were available including computerized tomography brain. The prognosis for affected patients is poor, with high mortality rate; most affected children may die by the second decade of life, if appropriate supportive measures are provided [8].

CS patient frequently develops photosensitivity dermatitis that results in desquamation and scarring of the skin. Our patient suffered from actinic chelitis of lower lip, which may be because of excessive exposure to the sunlight. A prematurely aged metabolic state has also been hypothesized as one of the principal causes of CS [9].

The CS can be diagnosed prenatally by examining amniotic cells cultured in vitro. The prenatal test can be carried out two to four days after the culture of sufficient cells [10].

CONCLUSION:
Anomalies of craniofacial and oral region are common in CS, and also life expectancy for the individuals suffering from this syndrome is comparatively short. Dentist should play a key role in the early diagnosis and management of oro-dental anomalies. Frequent examinations and emphasis on preventing dental disease must be stressed to the parents because of the difficulty in providing restorative care.

Appropriate and safe dental care for patients with Cockayne’s syndrome can be rendered after medical consultation.
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ORAL MYIASIS – A CASE REPORT

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ABSTRACT:

Oral myiasis is a rare disease caused by larvae of certain dipteran flies. A case of oral myiasis caused by chrysomya bezziana, in the maxillary anterior region in a 20 year old mentally challenged female patient is reported. Manual removal of the larvae by topical application of turpentine oil, surgical debridement of the oral wound, followed by oral therapy with broad spectrum antibiotics were used to manage the patient.

Key words: Oral myiasis, Chrysomya bezziana
(Submitted May 2012; Accepted June 2012)

INTRODUCTION:

The term myiasis is derived from Latin word ‘muia’ which means fly and ‘iasis’ means disease. The term was coined by F.W.Hope in 1840 [1]. Myiasis may also be defined as the infestation of live human and vertebrate animals by dipterous larva, which at least for certain period feeds on dead or living tissue of the host, liquid substances or ingested food [2]. It frequently occurs in rural areas infecting livestock and pets such as dogs and cats [2]. It has been reported in some unhealthy individuals in third world countries [3]. Incidence of oral myiasis is comparatively less than that of cutaneous myiasis because oral tissues are not permanently exposed to external environment [4]. Some cases of oral myiasis have been reported to occur following dental extractions, nasocomial infection, in drug addicts, in psychiatric patients [5, 6] and conditions that cause prolonged mouth opening like mouth breathing during sleep, senility, alcoholism and mental retardation [7]. The flies are attracted to the oral cavity due to neglected
oral hygiene or accumulation of fermented food debris [5 -7].

Persistent mouth opening facilitates the deposition of the eggs by the adult fly [5]. Subtropical climate and poor oral hygiene are conducive for breeding of the fly [8].

CASE REPORT:
A 20-year-old mentally challenged female reported to the out-patient with swelling of upper lip and left side of the face and presence of worms in the mouth from past 2 days. The patient was accompanied by her parents. The patient was of low socio-economic status, poorly built and apprehensive.

Extra oral examination revealed diffuse swelling of upper lip and left maxilla. Intra oral examination revealed necrotic area in the maxillary anterior region involving the vestibular sulcus in the maxillary central incisor region, measuring about 2.0 cm × 1.0 cm (Fig- 1); maggots were seen in the necrotic area. Based on the history and presence of maggots, a provisional diagnosis of oral myiasis was made. Cotton bud impregnated with turpentine oil was placed in the necrotic area for approximately 10 minutes. About 10-12 maggots were manually removed with tissue holding forceps and taken for entomological examination.

The area was then washed with saline followed by irrigation with betadine.

Broad spectrum antibiotics amoxicillin with clavulinic acid and ibuprofen were prescribed. The same procedure was repeated for two more days. Edema had considerably reduced after two days. Maggots were tapered in shape, creamy white in colour their segments giving the appearance of transverse rows, with brown black tip anteriorly and were identified as larvae of Chrysomya bezziana (botfly) by an entomologist. The patient was reviewed after two weeks and the results were satisfactory (Fig 2).

DISCUSSION:
Myiasis is caused by member of Diptera fly family that lays egg on food, necrotic tissue, open wounds and unbroken skin or tissue [9]. Chrysomya bezziana, the old world screwworm fly, is the cause of obligatory myiasis. The species is widely distributed throughout South-East Asia, China, Indian subcontinent, tropical Africa, and Papua New Guinea [10 -12]. Human myiasis due to Chrysomya bezziana is very rare; it was first reported in Hong Kong in 2003 [12].
The adult female lays egg on live mammals. The site of infestations is usually superficial wound, open sores and mucous membranes in the body orifices such as mouth, ear and nose [13]. The eggs hatch within 24 hours and the resulting larvae burrow into the host's tissues head downwards into the wound in a characteristic screw-like fashion, feeding on living tissue. The larvae release toxins to destroy the host tissue. The larvae complete their developments in 5-7 days, after which they then wriggle out of the wound and fall to the ground to pupate [3, 6, 14].

The standard treatment consists of topical application of turpentine oil, mineral oil, chloroform, ethyl chloride or mercuric chloride that make the larvae to come out followed by manual removal of the larvae and surgical debridement [5]. Systemic therapy of Ivermectin given orally in just one dose of 150-200mg/kg body weight and repeated after 24 hours has been found to be effective in several cases. It acts by blocking the nerve endings through the release of gamma amino butyric acid (GABA) leading to palsy and death of the parasite [12, 15]. The cases of oral myiasis with no medical systemic complications recover completely on removal of the larvae [16].

Infestations with chrysomya bezziana differ from other maggot infestations because there is tissue invasion even in absence of any pre-existing necrotic tissue. The Chrysomya bezziana maggots may cause serious and permanent tissue damage and extremely infested wounds can even lead to death of the host in absence of proper treatment. It is usually the people with mental and physical disability who are affected because of the difficulties in maintaining good oral hygiene due to poor manual dexterity [5]. Special care needs to be taken by the parents/guardians of
these patients as they are unable to maintain their basic oral hygiene.

CONCLUSION:
As the old saying goes “prevention is better than cure”, the disease should be prevented by controlling fly population, maintaining good oral and personal hygiene such as reducing the decomposition odour, cleaning and covering the wounds and by educating the susceptible population where basic sanitation is meagre. The personal taking care of special people is advised to ensure personal hygiene and adopt suitable practices to prevent the occurrences of infestations.

REFERENCES:
BILATERAL RECURRENT SPECKLED LEUKOPLAKIA: A CASE REPORT

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ABSTRACT:

Largely oral cancers are preceded by potentially malignant lesions, which may appear as white or red patches on the oral mucosa. Leukoplakia is one of the most common epithelial precursors of oral squamous cell carcinoma. Speckled leukoplakia is a rare type of leukoplakia with a very high risk of premalignant growth and mortality rate. Though it is the common precancerous lesion, it poses a major diagnostic and therapeutic challenge. We present a rare case of bilateral recurrent speckled leukoplakia with malignant transformation and discuss this relatively rare entity in light of current information from the literature. We also attempt to present the clinical relevance, and the therapeutic modalities available for the management of the disease.

Key words: Speckled Leukoplakia, White Patch, Tobacco Chewing, Precancerous Lesions

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INTRODUCTION:
Oral leukoplakia is characterized by adherent white plaques or patches on the mucous membranes of the oral cavity, including the tongue [1, 2]. The World Health Organization (WHO) currently employs the term Speckled Leukoplakia (SL) to describe the presence of both white and red patches on the oral mucosa [3, 4]. SL is a rare, highly aggressive, clinicopathological entity, with high-risk of malignant transformation and a precursor lesion of squamous cell carcinoma [2, 3]. Although the prevalence of SL in India is low, it presents histopathological features ranging from epithelial dysplasia to invasive carcinoma. This justifies placing these lesions among the oral lesions with the highest malignant potential.

CASE REPORT:
A 58 year old male patient reported to the department of oral medicine and radiodiagnosis, with the complaint of painless white patch on right and left buccal mucosa since 4 months.
Patient had a habit of cigarette smoking, 5 to 7 per day for a period of 30 years and presently patient had reduced smoking to 3 per day. He also had a habit of chewing unprocessed tobacco since 10 years with a frequency of 2 to 3 times a day. The content of the quid was betel leaf, areca nut, slaked lime, and tobacco. Along with these the patient also consumed alcohol occasionally.
Intraoral Examination of right and left buccal mucosa revealed firm, non tender, non scrapable, red and white patches measuring 2 x 2 cm. Surface appears rough and slightly elevated which clinically resembled “mud crack” in appearance. The lesion was not disappearing on stretching (Fig. 1).
Chair side investigation, toluidine blue staining was carried out to select the area of biopsy to be made. The selected area was then biopsied (incision).
Histopathologic examination with H and E stained sections showed hyperkeratotic stratified squamous epithelium overlying a fibrous connective tissue. Epithelium was showing hyperplasia with moderate to severe dysplastic features. Dense inflammatory infiltrate were also seen. The overall clinical and histopathological findings were considered diagnostic for SL with moderate to severe dysplasia. The patient underwent complete surgical removal of the lesion along with silicon graft (Fig 2). Post operative healing was uneventful (Fig 3). Patient was asked to visit the hospital for a regular check up once in a month.
Fig. 1: Red and white patch on right buccal mucosa

Fig. 2: Surgical excision on right buccal mucosa

Fig. 3: Healed surgical site after 3 weeks of surgery

Fig. 4: Recurrence of speckled leukoplakia after 3 months

Fig. 5: Histopathological photograph showing moderate to severe dysplasia
On 6 months follow up a recurrent lesion on the same buccal mucosa were seen. The lesion on the buccal mucosa was now showing more red areas on the surface (Fig. 4). They were diagnosed as SL. Both the lesions were taken for excisional biopsy. The H and E stained sections of both the lesions showed moderate to severe dysplastic features (Fig. 5). The patient is still under periodic follow-up once in every three months.

DISCUSSION:
WHO defines leukoplakia as a whitish patch or plaque that cannot be characterized, clinically or pathologically, as any other disease and which is not associated with any other physical or chemical causative agent except the use of tobacco [5]. The literature, however, strongly indicates the role of alcohol, viruses and systemic conditions [6, 7]. Various designations have been used to describe the presence of both white and red patches. Lesions appearing completely red are named as Erythroplakia. SL is indicated when red and white patches are present over the mucosa [2, 3]. WHO [4] currently recommends the term SL to describe mouth lesions that present red and white components; hence this term is used in the present case report.

Men are more commonly affected and the mean age at the time of diagnosis is slightly over 60 years [8, 9, and 10]. The buccal mucosa and tongue are the most common sites associated with SL; palatal mucosa, alveolar mucosa, gingiva, floor of mouth, and lip show a lower incidence. Typically, multiple oral sites can be also affected [11, 12], as seen in the present case.

Our case highlights the typical slow-growing, progressive, and persistent clinical course of this rare condition. As presented in our case, the initial finding can be an elevated non homogeneous whitish-grey lesion that tends to recur and proliferate, often over a protracted period of time, to result in a diffuse and widespread erythematous plaque that was associated with severe dysplasia.

Non-homogeneous leukoplakias are often associated with mild complaints of localized pain or discomfort and invariably develop into malignancy. Approximately 80 percent of SL progress to oral carcinoma over a period of time in spite of a variety of interventions. This feature contrasts with homogenous leukoplakia in which approximately 5-10 percent will transform into a carcinoma [2]. SL is resistant to most of the available treatment modalities, including surgery [4]. Therefore, total excision with free surgical margins is critical combined with a lifelong follow-up [4].

Malignant potential of leukoplakia is higher in women (6%) than in men (3.9%). Leukoplakia associated with habit of chewing tobacco shows higher rate of malignant transformation as compared to others [15]. In buccal mucosa and commissure region 1.8 percent malignant
transformation can occur. In lip and tongue region 16 to 38.8 percent malignant transformation has been reported. The annual malignant transformation rate has been determined to be 0.1% to 17% [16]. Less than half (33% to 42%) of leukoplakias undergo malignant change [17].

MANAGEMENT:
The degree of epithelial dysplasia plays a pivotal role while deciding the nature of treatment to the patient. Martorell- Calatayud [18] defined two risk groups and the subsequent treatment options:

Group 1: Those with low risk of malignisation:
Those leukoplakias lacking dysplasia, and those that show mild dysplasia located in low risk areas or those with a thickness of less than 200 mm or that present clinically as homogenous leukoplakia. A range of therapeutic approaches can be taken in this group: Regular patient follow-up. The interval between follow-up visits should not exceed 12 months in order to detect any change, suggestive of malignant transformation.

Treatment of lesions with topical or oral retinoids {eg: 13-Cis-Retinoic Acid (1.5 to 2 mg/kg body weight for 3 months) [18].

Treatments using nonsurgical ablative techniques, such as cryotherapy and carbon dioxide laser ablation. Of these options, the use of laser light has shown better results in terms of controlling the lesions, and so it is considered the treatment of choice in this low risk group.

Group 2: Those with high-risk of malignant transformation, which comprises:
Those leukoplakias with mild dysplasia located in high-risk areas measuring more than 200 mm, or those associated with a nonhomogenous clinical form; Leukoplakias with moderate or severe dysplasia; Verrucous leukoplakias. In this group, aggressive surgical treatment, consisting of excision of the entire thickness of the mucosa at the site of the leukoplakia is recommended. This is similar to the present case. Among the many therapeutic options available, however, eliminating risk factors (smoking, alcohol) and etiological factors (sharp broken teeth, faulty metal restorations and metal bridges) are preventive measure applicable to all patients with these lesions [13]. Regular check-up of these patients is essential, probably every 3, 6 and then 12 months, both in treated and untreated patients.

CONCLUSION:
Dentists, Dermatologists, ENT specialists, or Physicians may encounter asymptomatic initial and progressive lesions of leukoplakia on routine clinical examination. Patients with SL are benefitted if we have the earliest possible diagnosis and treatment, thus improving their prognosis. All the patients with a recurrent white lesion require vigorous follow up and have to be treated with aggressive approach.
REFERENCES:

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