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Spontaneous Autoamputation and Expulsion of Intestinal Lipoma

Sir,

Lipoma is a benign innocent tumour composed of fat, occurs predominantly in men. It is commonly found in subcutaneous tissue of neck, shoulder, back and buttock, occasionally it grows from the mesenteric and retroperitoneal fat and rarely from submucous coat of stomach and intestine, where it may form a polypoid mass.1

55 years old male presented with slight discomfort followed by mild vague pain in lower abdomen during night and passed a soft mass of fist size in the morning along with fecal matter (Fig. 1). No history of melana or fresh blood in the stool. 12 years back he had similar symptoms in the night and passed similar mass in the morning along with stool. He is socially alcoholic and nonsmoker. On examination, pulse-80/ min, BP - 110/78 mm of Hg. There was no subcutaneous swelling over the body. Abdomen and other systemic examination was unremarkable. Complete blood count, urine analysis, blood sugar and liver function, renal function and lipid profile were normal. Ultrasonography of abdomen and plain X-ray abdomen were normal. Upper GI endoscopy, colonoscopy, Ba meal follow through studies, plain and contrast CT abdomen did not reveal anything. On macroscopic examination, the mass was an encapsulated, circumscribed, lobulated of size 8 cm x 5 cm x 3 cm (Fig. 1) and on cut section, greenish yellow without any area of necrosis or haemorrhage. On microscopic examination, a circumscribed, encapsulated lesion composed of benign mature fat cells and focal areas of haemorrhagic infaction, possibly leading to autoamputation, thus histopathology suggestive of amputated polypoid lipoma.

Lipoma is a benign tumour of the gastrointestinal tract. It may occur in any part of small intestine but common in terminal ileum, usually submucosal and present as intraluminal single polypoid mass without any symptoms, but once the mass lesion exceeds 4 cm in diameter, it may present as lead point of intussusception or cause of obstruction on laparotomy and rarely bleeding may occur from the over lying mucosa.2 Ba meal follow through study has detection rate of 50% to 70% of small bowel tumours. Further enteroclysis has been considered useful for demonstrating ileal tumours and Ba meal follow through examination for proximal bowel. Still further, conventional Ba meal follow through examination is superior to CT abdomen for evaluating intramural and mucosal disease, whereas CT abdomen is more accurate for evaluating intramural and retroperitoneal masses. Submucosal lipomas on Ba meal follow through studies typically demonstrate, sharply defined mobile intramural sessile or pedunculated defects, which changes with the compression.3

This time, reported case was evaluated thoroughly and no other soft mass was detected throughout the gastrointestinal tract, but definitely twice with an interval of 12 years a polypoid lipomatous mass is expelled, which might be due to spontaneous autoamputation. Available literature was reviewed, such type of spontaneous autoamputation and spontaneous expulsion of polypoid lipoma from gastrointestinal tract is not reported so far.

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Hodgkin’s Disease and Pilocytic Astrocytoma-A Synchronous Presentation

Sir,

Secondary neoplasm after treatment of Hodgkin’s disease is a well-established entity but synchronous neoplasm associated with either Hodgkin’s disease or astrocytoma is very uncommon.

A 16-year-old boy presented in OPD with right-sided neck swelling for 5-6 months. There was no history of fever, weight loss or night sweats. On examination, patient had right cervical lymph node 4x3 cm, three in number, discrete rubbery and mobile. No other lymph nodes were palpable. There was no organomegaly. Clinical Examination of other systems was normal. Patient underwent a lymphnode biopsy, after routine baseline tests. Next day he developed generalized tonic clonic

Fig. 1 : Soft polypoid mass.
seizure. There was no focal neurological deficit and ophthalmologic examination was normal. CT scan brain was done which showed space occupying lesion in cerebellum. Sterotactic biopsy was done from that site, which showed pilocytic astrocytoma. Lymph node biopsy was reported as Hodgkin’s disease nodular sclerosis. Hemogram, biochemistry, x-ray chest, ultrasound abdomen and bone marrow aspiration were normal. The plan of treatment was discussed with the radiation oncologist for the feasibility of radiotherapy treatment for both the diseases. Due to toxicity radiotherapy as a single modality was deferred. Patient was started on C-MOPP (cyclophosphamide, vincristine, procarbazine, prednisolone) and radiotherapy was given to the brain. Patient received six cycles of chemotherapy and 50 Gy of radiotherapy in 25 fraction. His disease is in complete remission for one year. Secondary neoplasms after treatment of Hodgkin’s disease are common. They may occur years after the treatment. Synchronous malignancies with Hodgkin’s disease or pilocytic astrocytoma are extremely uncommon. Non-Hodgkin lymphoma, multiple myeloma, carcinoma breast and thyroid has been occasionally reported as synchronous malignancies with Hodgkin’s disease.1

Synchronous malignancies with astrocytoma are carcinoma lung, uterus, stomach and breast, which has been rarely reported only with high grade astrocytoma (Glioblastoma multiforme).2

Pathogenesis of multiple primary neoplasia is not known. Intrinsic factors like heredity, age, and immunity have been proposed as possible mechanisms. Extrinsic factors such as exposure to environmental radiation and chemicals may also be important.2

Management decision for these patients are crucial. Treatment delay could lead to poor outcome in cancer patients. In our patient considering the treatment outcome and possible toxicity, chemotherapy for Hodgkin’s disease and radiotherapy for astrocytoma were started simultaneously.

One case report with glioblastoma multiforme of brain stem associated with Hodgkin’s disease has been reported.3 As per our knowledge this is the first case report of Hodgkin’s disease synchronously presenting with low grade astrocytoma.

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Torsade De Pointes Probably Induced by Sparfloxacin


I have read the above case report with great interest. I would like to offer the following comments:

The direct involvement of heart in patients with tuberculosis resulting in arrhythmias/sudden cardiac death is rare, but well known complications of tuberculosis. In fact, prolongation of QT interval due to cardiac tuberculosis has also been reported in literature. In the present case, this possibility also needs to be considered. Therefore, sparfloxacin as the only cause of a prolonged QT interval needs to be reconsidered by the authors. The only approach to resolve this dilemma would have been to perform an autopsy. In the present case, it would have provided us with valuable information about the exact cause of death in this young female.

Unfortunately, in our country due to the socio-cultural limitations, we would all agree it is almost impossible to get consent from the relatives to perform even a limited autopsy. At the same time, the importance of an autopsy is also not fully appreciated by the medical fraternity. The need to educate the public as well as the health care workers about the benefit of this procedure in the progress of medical science should be a matter of urgency for our fraternity in India. In fact, the same stigma is also responsible for the failure of organ transplantation programme in our country in the few centers it was initiated.

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Reply from the Authors

Sir,

We appreciate the thought provoking diagnosis suggested by Dr. Ahluwalia. Myocardial infarction is a rare condition which we did not consider in the differential diagnosis as our patient had no features of myocarditis.

Grossly myocardial presentation has three types, tuberculomas, military tuberculosis as a complication of generalized military disease and lastly as diffuse infiltrative disease associated with pericarditis. The myocardium is usually affected by direct extension from lymph nodal disease or less often by retrograde lymphatic drainage from tubercular mediastinal lymphadenitis. Military infection may spread via blood and direct spread occurs from pericardium. There was no mediastinal lymph nodes or any evidence of military or pericardial disease to suggest extension onto myocardium.

We agree with the suggestion of autopsy but such facilities are not available in big centers of our country.

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Air Pollution

Sir,

Recent review article (JAPI 2002;50:583-7) on association between air pollution and allergic diseases is timely important. With reference to the article, we have provided some more information below.

a) Indian scenario on hazards of air pollution on respiratory system, eye and pregnancy and the need for multisectroial approach to combat the malady was highlighted earlier. According to the 1998 National Ambient Air Quality Monitoring Programme (NAAQMP) data, the concentration of particles less than 10 micron (PM 10) levels in the residential areas of Delhi are critical.

b) In India NAAQMP was initiated in 1981 and currently NAAQMP has net work comprising of 220 stations covering 92 cities/towns. Central pollution control board (CPCB) brings out national data through their publication and website (<www.envfor.nic.in/cpcb>). Apart from that many other reports as well as publications are brought out by National Environmental Engineering research Institute (Indian Journal of Environmental Health and Journal of Indian Association for Environmental Management), Nagpur 400 020.

c) India has become one of the few countries of the world to enshrine in its constitution, a commitment to environmental protection and improvement, and hence, introduced comprehensive set of laws. However, implementation requires political will, unstinted efforts of authorities, public support, cooperation from religious leaders, teachers and non-governmental organisations, etc.

d) Role of Indian apex court: In the sensational ‘CNG case’, a three judges bench headed by BN Kirpal of the apex court (supreme court) of India, while indicating the centre, drew its attention to the increase in respiratory diseases due to air pollution. As per a world bank study, the annual cost incurred by India on health was upto about Rs.5500 crores (approximately over US$one billion) due to ambient air pollution and out of this the cost for Delhi alone was found to be about Rs.1000 crores, (0.19%), the
bunch said, adding, that alternative fuel of CNG, LPG and electricity was a preferred technology which critically polluted cities like Delhi need as a leap frogging technological option. Role of adulteration of vehicle fuel and its effect on air pollution was also mentioned in the recent supreme court judgement (The Hindu, Madurai 7th April, 2002). Healthcare professionals and the public should appreciate the strong words of the apex court, which reflect their concern over the health of children, the wealth of the nation. The apex court has rescued the public when professionals and authorities have failed to execute their responsibilities.

Physicians, being responsible citizens of the nation, should take efforts through professional associations and public awareness campaigns to bring down/control pollution or else we have to carry oxygen mask and cylinder like people carrying drinking water everywhere in India.

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Paroxysmal Nocturnal Movements Secondary to Obstructive Sleep Apnoea Syndrome

Sir,

A middle aged man manifesting with involuntary movements of the head, limbs and trunk soon after falling asleep is described. He also had snoring, feeling of unsatisfactory sleep on awakening, apnoea during sleep and excessive daytime somnolence. He was obese and had large neck circumference, narrow oral orifice, large tongue, prominent uvula, low palatal arch and enlarged tonsils. Video telemetric EEG documented involuntary movements and excluded seizure phenomenon. Polysomnography confirmed obstructive sleep apnoea. Correlation was noted between abnormal movements, arousals and the episodes of apnoea and subsequent oxygen desaturation. We believe that these involuntary movements are a rare manifestation of the obstructive sleep apnoea syndrome.

Obstructive sleep apnoea syndrome (OSAS) is a common but under-recognized cause for dysomnia and is potentially treatable. The essential features of the disorder are well known and restlessness with involuntary movements of the head, trunk and limbs are also described. However, abnormal movements as the presenting manifestation of OSAS is rare. We describe a patient who reported with paroxysmal nocturnal movements of several years duration to neurology services and on evaluation was detected to have OSAS.

A 48 years man presented with episodic abnormal movements of the head of 18 years duration. Each episode would begin soon after going to sleep and consisted of involuntary, jerky, no-no movements of the head and tight closure of the eyes. He could understand commands during the latter part of episodes but was not able to respond. All episodes were of similar nature and were preceded by headache. After the initial disturbance, subsequent sleep would be free of these movements. Temporal association with tensions and stress was observed and such situations were avoided by him. The movements also caused considerable strain on the marital relationship. Severity and frequency of movements increased over the two years prior to consultation at our institute and other movements such as head banging, tonic posturing of the head, hyperextension of neck, opisthotonic posturing of the trunk and abnormal posturing of fingers appeared. In addition to this, patient had excessive daytime sleepiness (EDS) that required change of occupation from driving to painting. He scored 18 on the Epworth Sleepiness Scale.

On waking up he often noticed tiredness and headache. He was a snorer for 25 years with recent worsening. During sleep, family members often noticed periods of apnea. Sleep hygiene was satisfactory. He was not an alcoholic or a smoker. He had been under the care of several practitioners and specialists for these complaints for 18 years with varying diagnoses including dissociative motor disorder, paroxysmal nocturnal movement disorder and adverse seizures. Polypharmacy was the rule, resulting only in side effects. On examination, his body mass index was 30 kg/m² and adjusted neck circumference (neck circumference + 3 cm for snoring) was 51 cm. He had noisy breathing, prominent inferior turbinates of the nose, narrow oral orifice, large tongue, prominent uvula, low palatal arch and enlarged tonsils. There was no evidence of cor pulmonale or congestive cardiac failure. The movements were confirmed to be sleep-related by direct observation, video telemetric EEG (Telefactor) and polysomnography (32 channel Neurofax EEG 2110 Nihon Kohden machine with Polysmith software). Sleep efficiency was 19%. Apnea-hypopnea index was 120 per hour and longest respiratory disturbance lasted 53 seconds. Oxygen saturations were as follows: average - 86%, minimum - 52% and during longest episode of apnoea - 70%. There were 55 arousals during the recording period. The abnormal movements were observed during initial non-rapid eye movement sleep. Correlation was noted between abnormal movements, arousals/awakenings and the episodes of apnea and subsequent oxygen desaturation (Fig. 1). There were no epileptiform discharges. Magnetic resonance imaging (MRI) demonstrated obstruction in the oropharynx, more so during sleep (Fig. 2). Pulmonary function tests showed mild restrictive
pattern. It was concluded that the patient had restless sleep due to behavioural arousals, secondary to obstructive sleep apnea syndrome. Recent worsening was attributed to the use of benzodiazepines.

The causes for abnormal movements in sleep include dyssomnias such as periodic limb movement disorder and parasomnias such as head banging, sleep starts, nocturnal paroxysmal dystonia, frontal lobe seizures and rapid eye movement sleep behavior disorder. Stereotypic shaking of the legs, distinct from periodic limb movements in sleep, has been reported in a patient of OSAS. Through a detailed history, observation of the events and polysomnography, all these diagnoses were excluded and a diagnosis of obstructive sleep apnea was established.

Forty four percent of middle-aged men and 28% of middle-aged women snore habitually. When diagnostic criteria for OSAS is taken as apnoea-hypopnoea index of five or more plus EDS, 4% of men and 2% of women in middle-age are affected. Thus it is an important public health challenge whose prevalence is more than many of the out-patient neurological illnesses such as epilepsy. It is the single most common disorder in sleep centres and is responsible for more morbidity and mortality than any other sleep disorder. Inspite of its high prevalence, 99% of people with OSAS go undiagnosed.

Primary snoring and OSAS represent two ends of the spectrum of sleep-disordered breathing resulting from narrow upper airways. Normally, negative intrathoracic pressure results in suction of the oropharyngeal tree and its subsequent narrowing. However the pharyngeal dilator muscles reduce the tendency for collapse by a negative pressure reflex. In sleep, this reflex is attenuated leading to increased upper airway resistance. In patients of OSAS, pharyngeal dilator

Fig. 1: Episodes of oxygen desaturation and apnoea during sleep recording

Fig. 2: Fast spin echo T1W1 in parasagittal plane. (A) With patient awake showing the large tongue almost filling the oral cavity, (B) With patient asleep showing the oropharynx obliterated by the apposition of uvula with pharyngeal wall and large tongue opposing the palate
muscle activity is less and in addition, tissue pressure compromises pharyngeal patency. Thus upper airway resistance is significantly increased, leading to obstructive apnea. This may result in hypoxia and hypercarbia and arousal to lighter stages of sleep or even awakening. As a result, the tone of the pharyngeal dilator muscles increases and patency of the airway is reestablished.2,7 Such EEG arousals may be accompanied by behavioral arousals such as positional change, upward raising of the torso off the bed and large flailing movements of the limbs. Patients may appear agitated and there may be crying or groaning. Only rarely do patients recall these arousals. This leads to sleep fragmentation and EDS.2 It is unusual for behavioral arousals to be the presenting features of OSAS as was observed in our patient. Benzodiazepines reduce pharyngeal dilator tone and its use by the patient may explain recent worsening of the involuntary movements. In view of manifest EDS, multiple sleep latency test (MSLT) was not necessary.2 Treatment of OSAS includes lifestyle modifications, pharmacotherapy using protryptilline, nasal continuous positive airway pressure (CPAP) or bilevel positive airway pressure (BiPAP) appliances and surgical techniques like uvulopalatopharyngoplasty (UPP) and geniotubercle advancement,2,7,8 or a combination of these. In view of the high apnoea-hypopnoea index and profound oxygen desaturations, nasal CPAP is being planned for our patient.

Clinical vigilance and detailed polysomnography in other patients with sleep-related movement disorders may expand the spectrum of obstructive sleep apnoea syndrome and facilitate treatment to alleviate morbidity.

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