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Dr. Tejaswi P
III Year Postgraduate
Student, Department of
Pathology, Kamineni
Academy of Medical Sciences
and Research Centre, LB
Nagar, Hyderabad, Telangana,
India

Dr. Shailaja Prabhala
Professor, Department of
Pathology, Kamineni
Academy of Medical Sciences
and Research Centre, LB
Nagar, Hyderabad, Telangana,
India

Dr. Ashok Kumar Deshpande
Professor and Head,
Department of Pathology,
Kamineni Academy of Medical
Sciences and Research Centre,
LB Nagar, Hyderabad,
Telangana, India

Corresponding Author:
Dr. Shailaja Prabhala
Professor, Department of
Pathology, Kamineni
Academy of Medical Sciences
and Research Centre, LB
Nagar, Hyderabad, Telangana,
India

Lipoma: A ubiquitous tumor at uncommon locations

Dr. Tejaswi P, Dr. Shailaja Prabhala and Dr. Ashok Kumar Deshpande

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Abstract

Lipomas are the most common mesenchymal neoplasms in the body. Most common locations being head, neck and extremities. Other very rare locations are tongue, intestine, buccogingival junction, parotid, lips and uterus. We have presented three rare sites of lipoma of which first is a case of 75 year old male who presented with problems of mastication which on examination showed soft swelling on the left lateral tongue and later the diagnosis of lipoma was confirmed. Next is a case of intestinal lipoma where the patient presented with intestinal obstruction. MRI showed jejuno-ileal intussusception. Here, Submucosal lipoma was confirmed on histopathology. The last case is of a 61 year old female who presented with post menopausal bleeding, MRI was suggestive of fibroid uterus with fatty signal, which was confirmed as an intramural lipoma on histopathology.

Keywords: Lipoma, ubiquitous tumor, uncommon locations

Introduction

Lipomas are the most common benign neoplasms of mesenchymal origin and may arise in any location ^[1]. They are frequently found in the head and neck regions in about 13% of the cases. The uncommon sites of presentation of lipomas are in the parotid, intestine, tongue, uterus, buccogingival junction and other sites.

Case 1: A 75 year old male presented to the oncology OP with a swelling on the left of the tongue since 2 months. Patient also complained of masticatory problems since 1 month. Patient is known case of hypertension and was taking medications the same. No other medical co-morbidities were noted. Patient gives a past history of road traffic accident with multiple rib fractures and tongue laceration where the present is situated. On examination, the swelling was non tender. Local examination showed a swelling of size 2cm seen on the lateral border of tongue. The surrounding mucosa was normal, no redness or ulceration was noted. The free border of the tongue showed yellow areas. The regional lymph nodes were not palpable. With the above clinical and examination findings was lipoma as the first differential diagnosis. MRI was done outside for which images were not available. The report showed a well circumscribed lesion within the muscles of tongue in the anterior two-thirds area on left lateral side. Diagnosis was made as lipoma. Planned for local excision, the surgical profile was done which was normal. Local excision with a surrounding rim of normal tissue was excised under general anesthesia. The excised area was reconstructed by vicryl sutures. The patient was discharged on the 5th day with postoperative care instructions.

Gross: Well circumscribed soft mass which on external examination was on the superior side covered with mucosa. The specimen was sent to the pathology department for further examination which was labeled for margins. Cut section revealed a well delineated, non encapsulated, yellowish, solid, greasy mass measuring 1.8×1.7×1cm. The grossed bits was taken for routine histopathological processing.

Micro: Stratified squamous epithelium with underlying lesion composed of mature adipocytes was noted. One section also showed entrapped muscle fibers. Inflammation, atypia, necrosis or lipoblasts were absent. Therefore a diagnosis of intramuscular lipoma of tongue was made. (H&E, x40)

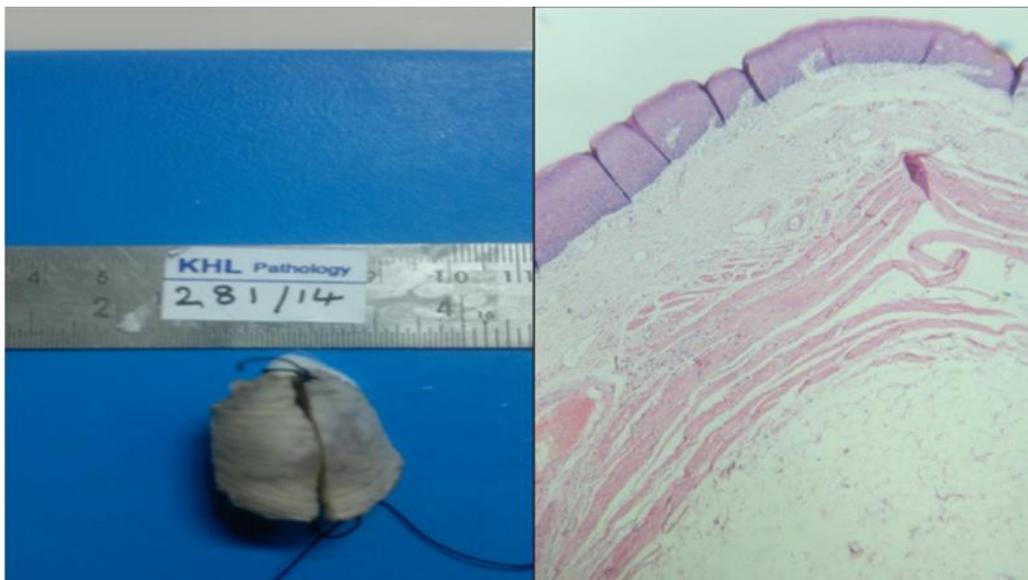


Fig 1a: Shows the excised lesion covered by mucosa. **1b:** Stratified squamous epithelium with lobules of mature adipose tissue and entrapped skeletal muscle

Case 2: A 37 year old male presented with complaints of abdominal pain and multiple episodes of bilious vomiting. He had fever with chills of 4 days duration associated with generalized body pains. There was no history of loose stools/constipation/burning micturition. No other comorbidities were noted. On examination patient was conscious and oriented. Vitals were stable. Per abdomen was soft, distension was noted. Bowel sounds were faint to absent. Guarding and rigidity were noted. Clinical diagnosis of intestinal obstruction was made. CT showed jejuno-ileal intussusception. After a thorough surgical profiling patient was taken up for laparotomy+reduction of jejuno-ileal

intussusception+resection and anastomosis of small intestine under general examination.

Gross: A part of small intestine measuring 12cm. Mucosal surface showed three polypoidal masses with surface ulceration, largest measuring 3.5×2.5×2.5cm. Cut section of masses were yellowish, greasy.

Micro: Section showed small intestinal mucosa with submucosa having lobules of mature adipose tissue. (H&E, x40)

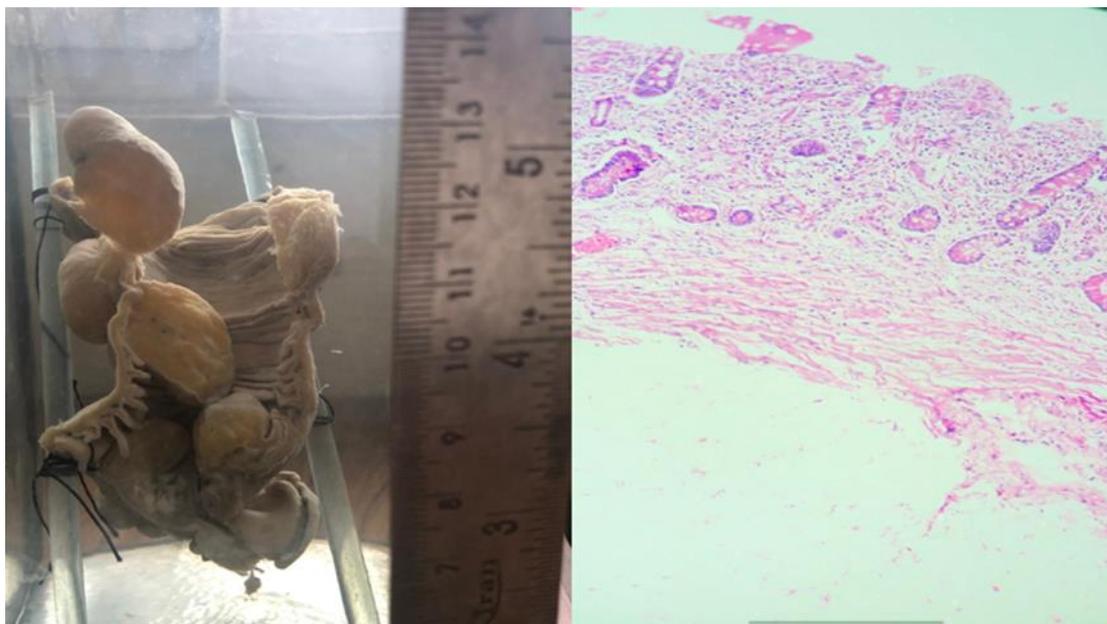


Fig 2a: Shows gross specimen of part of intestine with multiple well circumscribed yellow nodules. **2b:** Section shows unremarkable intestinal mucosa and the submucosa shows lobules of adipose tissue with intervening fibrous septa suggestive of Lipoma

Case 3: A 61 year old P3L3 presented with complaints of post menopausal bleeding (1 episode) one month back and sensation of mass per vaginum since one year. Her obstetric history: previous 3 normal vaginal deliveries and last child

birth was 28 years ago. She is a known case of diabetes mellitus and hypothyroidism and is taking the medication for the same. Menstrual history: attained menopause 15 years back. On admission: on examination, pulse rate was

82/min, BP: 110/70 mm of Hg. Systemic examination: CVS: S1, S2 heard, RS: BAE+, clear. Per abdomen was soft, non tender, obese. Per speculum: second degree uterine descent present, cervix showed circumoral erosion and Cystocele of grade 1. Per vaginum uterus was anteverted, size could not be made out, mobile, bilateral fornices were free.

Presurgical laboratory profile was within normal limits.

Chest X ray was normal. Pap smear was consistent with cervical erosion associated with acute inflammation, with moderate reactive atypia. Ultrasound abdomen showed increased endometrial thickness with heterogeneous echotexture. MRI showed bulky uterus with evidence of 2.3x2.7 cm well defined rounded fatty signal intensity, space occupying lesion in anterior myometrium indenting and displacing the endometrial cavity posteriorly. 1.9x1.5 cm well defined sub serosal fibroid in the posterior wall was also noted. Impression was given as multiple fibroids in uterus, largest in anterior intramural fundic region showing

fatty degeneration. Mild decent of uterine cervix into vagina was noted. After routine surgical profile, patient was taken up for total abdominal hysterectomy and bilateral salpingoophorectomy under spinal anesthesia. Specimen was sent for hisopathology. Her postoperative period was uneventful and patient was discharged with due advice.

Gross: Uterus measured 8x6x4cm. External surface of uterus was nodular with single nodular mass measuring 2.5x1.5 cm. The cut section showed cervix measuring 2.5cm, endometrium measuring 0.2 cm and myometrium measuring 2.2 cm. A single intramural well circumscribed nodule was noted which was homogenous yellow and soft in consistency. Attached right and left adnexae were nil-remarkable.

Micro: Showed lobules of mature adipose tissue suggestive of intramural Lipoma of uterus. (H&E, x40)

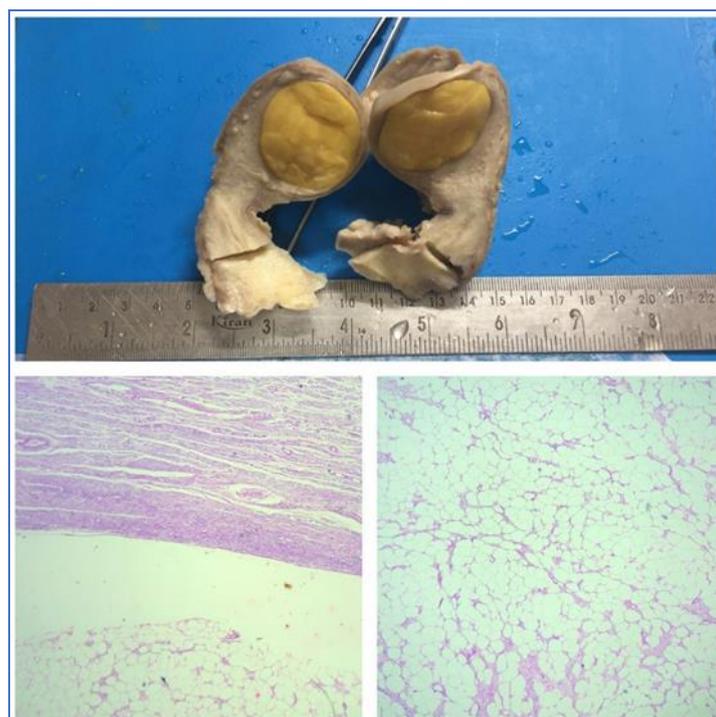


Fig 3a: Shows gross specimen of uterus with a well circumscribed nodular mass, **3b:** Shows uterine mucosa with lobules of adipose tissue, **3c:** Shows lobules of adipose tissue

Discussion

Lipomas are most common soft tissue tumors composed of well-differentiated adipocyte cells [1]. They are usually asymptomatic but may cause discomfort or pain with direct pressure. Lipomas can be present in any organ throughout the body. They constitute 15–20% of cases occurring in head and neck region [2]. Multiple rare sites for lipoma have been reported like tongue [3], bucco-pharyngeal, hard palate [4], parotid [5], uterus [6], plantar surface [7], intestine [8]. Intraorally, most commonly involved sites are buccal mucosa, tongue, floor of the mouth, buccal vestibule, and lips. Many syndromic conditions are associated with multiple lipomas. The occurrence of multiple lipomas can be seen in association with Cowden's syndrome or multiple hamartoma syndrome, Frohlich syndrome, Proteus syndrome, and Bannayan–Zonana syndrome. Most common mucocutaneous lesions in the patients with Cowden's

syndrome are small papular lesions in palate and gingiva, papillomatous and verrucous lesions of buccal mucosa, fissured tongue, and multiple cutaneous lipomas. Frohlich syndrome also known as prune belly syndrome is defined by multiple lipomas, obesity, and sexual infantilism. Proteus syndrome is marked by multiple lipomatous lesions, including pelvic lipomatosis, fibroplasia of feet and hands, skeletal hypertrophy, exostoses and scoliosis, and various pigmented lesions of skin. A case of congenital lipoma was described in a 7-year-old boy in upper labial frenum. Bannayan–Zonana syndrome is characterized by congenital association of multiple lipomas, hemangiomas, and macrocephaly.

Mean age of occurrence of intraoral lipoma varies according to different studies, but they usually occur in fourth and fifth decades of life. The occurrence of lipoma is uncommon in children. Their prevalence is similar in both the genders,

although a male and female predominance has also been recorded.

In this study, we are presenting 3 cases of rare locations of lipomas at three unique locations like the tongue, intestine and uterus.

The first case in this case report is a 75 year old with the swelling of the tongue. Patient presented with difficulty in mastication. Histopathology section showed stratified squamous epithelium with lobules of mature adipose tissue. Other section showed entrapped skeletal muscle. (figure 1b, 1c)

Lipoma of tongue accounts for only 0.3% of all tongue tumors. Roux gave the first description of an intraoral lipoma in 1848 and he referred to it as a "yellow epulis" [9]. Tongue which is totally devoid of fat cells is a rare site for lipoma. This is one such rare case of the universal tumor, presenting at the lateral margin of the tongue, for which complete tumor excision was done. The larger ones can cause macroglossia, atrophy of tongue musculature, dental abnormalities such as anterior open bite and masticatory difficulties as seen in this case, as well as airway and speech problems [3]. The symptoms also depend upon the secondary changes seen in the lipoma like the volume, texture, tenderness and ulceration on the lesion [10]. Oral lipomas clinically present as slow growing solitary or multiple lesions which may be sessile or pedunculated. Many other oral lesions mimic a lipoma on clinical presentation such as oral dermoid and epidermoid cysts, oral lymphoepithelial cyst, benign salivary gland tumor, mucocele, benign mesenchymal neoplasm, ranula, ectopic thyroid tissue, and lymphoma are considered in its differential diagnosis. Various theories have been thought for the pathogenesis of lipoma at the rarer sites like in a case of lipoma of tongue. First is "hypertrophy theory which occur due to obesity and inadvertent growth of fat. As this couldn't justify the cause of lipoma at sites devoid of fat alternate theory called the metaplasia where there is aberrant differentiation of normal mesenchymal cells to fat forming cells (lipoblasts) which in turn undergoes proliferation to give rise to lipoma. Triggering agents for metaplastic theory are trauma, chronic irritation that leads to proliferation of lipoma [2]. Proposed hypothesis is that after a soft tissue injury and hematoma formation there are various cytokines involved in the repair process which probably play a role in the preadipocytic differentiation and proliferation. [9] Similar explanation can be given to the case where she already gives a history of laceration of tongue [2]. Lipomas have been classified according to their morphological features into simple lipoma, fibro lipoma, spindle cell lipoma, angiolipoma, myolipoma, pleomorphic lipoma, myxoid lipoma, atypical lipoma and intramuscular lipoma [10].

The second-case report is the case of 34 year old with submucosal lipomas. Lipomas of the colon were first reported by Bauer in 1757 [12]. Several studies have shown that the most common site of colonic lipomas is the ascending colon (45%), although tumors may occur in the sigmoid colon (30.3%), descending colon (15.2%), and transverse colon (9.1%) [8]. Lipomas of the digestive tract are rare and most often found incidentally during a colonoscopy, computed tomography (CT) scan, surgery, or autopsy [12]. Lipomas of digestive tract are asymptomatic until they are more than 2 cm in size. Larger lesions may become symptomatic which cause a radiological confusion

with malignancy [8]. They are usually solitary with varying sizes and may be sessile or pedunculated [11].

Symptomatic patients present with abdominal pain, hemorrhage, diarrhea, or constipation. More larger lesions can lead to obstruction and intussusception requiring surgical or endoscopic resection. Radiologically, three endoscopic signs help in the diagnosis of lipomas due to their location mostly in the submucosa. Ninety percent of colonic lipomas are localized to the submucosa; colonic lipomas are rarely found in other layers of the bowel wall. Due to this location, three endoscopic signs can aid in the diagnosis of a lipoma: the "cushion" sign, which occurs when forceps press into the mass, resulting in a depression or pillowing; the "tenting" sign, which occurs when mucosa is grabbed over the lesion and pulled away, resulting in a tent-like appearance; and the "naked fat" sign, which occurs when fat is grossly extruded after biopsy. This submucosal position also aids in the treatment options by endoscopy like end loop excision, nylon loop assisted removal, endoclipping and sectioning of overlying mucosa [8]. Lipomas can also be seen in the subserosal or intramuscular planes rarely.

In the present study patient presented with intestinal obstruction which radiologically was a jejunoileal intussusception. On final histopathology sections revealed a intestinal mucosa with submucosa showing lobules of mature adipose tissue with intervening thin fibrous septa. (figure 2b, 2c)

The third case report is of a 61 year old female who presented with post menopausal bleeding. Pure lipoma of the uterus is extremely rare.

The incidence of uterine fatty tumors varies from 0.03-0.2% [13]. Pure lipomas can attain great size before coming to clinical attention and are mistaken for carcinomas due to the old age of the patient, rapid progression, and pressure effects [14]. These tumors commonly arise in postmenopausal women. Common predisposing factors are hyperestrogenism, hyperlipidemia, diabetes mellitus, hypothyroidism and toxemia in pregnancy. Pure uterine lipomas are at one end of the spectrum are composed entirely of adipocytes, on the other end it can be intermixed with various connective tissues or smooth muscle in varying proportions hence labeled as Lipoleiomyomas and Lipofibromas [15]. Majority are intramural. They can be subserosal or submucosal. These tumors are of unknown histogenesis and nomenclature. Histogenesis is controversial, most likely from pluripotent connective tissue cell. Other theories proposed are misplaced embryonic fat cells, metaplasia of muscle or connective tissue into fat cells, proliferation of perivascular fat cells, and fatty infiltration or degeneration of connective tissue [13].

Magnetic resonance imaging can be useful in identifying the fatty nature of the lesion as seen in our case where MRI showed fatty signal intensity.

Paraffin embedded sections stained with H&E showed uterine wall with the submucosa showing lobules of mature adipose tissue with thin intervening fibrous septa. Uterine lipomatous tumors are uncommon neoplasms and are categorized into: (1) Pure lipoma which is composed of only mature fat cells. (2) Lipoleiomyoma, angiolipoma, and fibromyolipoma which consists mixture of mature adipose tissue, smooth muscles, fibrous tissue, and other connective tissue elements. (3) Liposarcoma which is a malignant

neoplasm and immunohistochemistry (IHC) studies play an integral role in understanding its complex histogenesis. Immunohistochemical (IHC) stains such as smooth muscle actin (SMA) and vimentin can be done in sections of both the tumors to confirm the nature of cells especially in cases where they can be mistaken for malignancy especially in elderly women. Typically Smooth muscle cells, endothelial cells from the tumor of uterine corpus will show immunoreactivity with SMA whereas adipocytes will be negative. Vimentin also shows immunoreactivity with smooth muscle cells, endothelial cells and mature adipocytes^[14].

In our patient, total hysterectomy was done, and there was no evidence of gynecological malignancy. H&E could eliminate the diagnosis of lipomasarcoma and therefore IHC was not done. In view of complex histogenesis and association of gynecological malignancies and metabolic disorders in patients of uterine lipoleiomyoma, more large number case studies with IHC findings and follow-up are required to understand histogenesis.

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