Lymphangiomas are benign congenital malformations of lymphatic system characrerized by multiple communicating lymphatic channels and cystic spaces.

Cystic hygroma is most common variety of lymphangioma. Vast majority of lymphangiomas are diagnosed before the age of 2 years and manifest anywhere in the body but most frequently in cervico-facial region. These may extend from cervical region into axilla and mediastinum or into floor of mouth and tongue. These are delineated into microcystic and macrocystic forms. Though MRI is the diagnostic investigation of choice US is regularly used initialy to confirm the cystic nature of lesion. US feature includes multiseptated anechoic or hypoechoic lesions with vascularity along the wall and septae (macocystic forms) while it appears as hyperechoic mass in microcystic variety. Branchial cleft cyst and cystic hygromas have quite similar soft tissue characteristic on CT or MRI as well. However, cystic hygroma does not cause any displacement of structure in contrast to branchial cleft cyst. Non-surgical treatments include sclerotherapy, radio-frequency ablation or laser therapy may be applied while surgical resection may be considered as a last resort (10,11).

Reported case of 12 years old boy presented with a soft non-tender lateral neck swelling. His physical examination and US findings were not conclusive. Though the mass was extrathyroidal in origin but branchial cleft cyst and cystic hygroma are the common differential diagnosis. No CT pathognomic findings was available as well. Cytomorphological study may exclude malignancy but confirmation of diagnosis only possible by histopathological examination.

Cystic neck masses in children can be a diagnostic dilemma for which many investigative prodecures are used. However for initial imaging, ultrasound can be a first choice followed by MRI if neccesary. However histiopathology is the only method for definitive diagnosis.

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Juvenile Recurrent Parotitis: A Rare Case Report

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Juvenile Recurrent Parotitis (JRP) is a rare disorder in children characterized by recurrent non-suppurative inflammation of parotid gland. Such a case is presented here in which, a 6 year old girl presented with recurrent painful swelling in front of left lower end of mandible and was later diagnosed as a case of JRP. High resolution ultrasound evaluation of parotid gland paved the way to the diagnosis of such a rare condition.

Keywords: Parotid gland, Juvenile, Parotitis, Ultrasonography.

Juvenile recurrent parotitis (JRP), also known as infantile chronic recurrent parotitis (ICRP) is a rare, recurrent, nonobstructive and nonsuppurative parotid inflammation in young children with a multifactorial etiology (1,2). It was referred as the second most common salivary gland disease of childhood, after mumps in the time before the universal vaccination in infancy (3). It is a rare condition whose aetiology still remains elusive. It is usually unilateral, but bilateral exacerbation can also occur, recurring at least twice before puberty with eventual termination of the disease in the second decade of life (4).

CASE REPORT

A 6 years old girl visited Institute of Nuclear Medicine and Allied Sciences (INMAS), Rajshahi for high resolution ultrasound study (HRUS) of a painful swelling on her left lateral face below left ear. She had all together 7 such episodes in the last 2 years, each time the swelling lasted for about one week. For this she received homeopathic medications irregularly. The last episode lasted for 6 days. During these episodes she had low grade fever but had no oral dryness, increased salivation or altered taste. There were no history of dryness of eyes, joint pain or swelling and skin rashes. There was no family history of parotid swelling. On examination the child was afebrile with a small swelling located right below the left ear. It was tender on touch with no skin changes, smooth and soft in consistency. Right parotid region appeared normal.

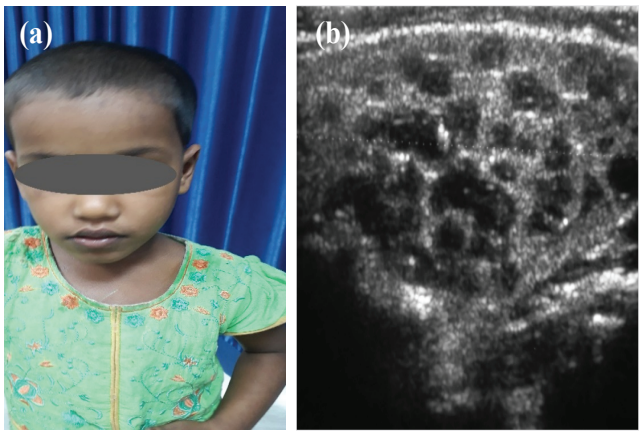

Figure 1: (a) Swelling in left parotid region. Figure 1 (b): Ultrasonographic image of left parotid gland showing enlarged heterogenous parotid gland (3.4 x 1.9 cm) with multiple small nodular areas.

Pressure over the gland did not cause any purulent discharge from the parotid duct. There was no sign of xerophthalmia or xerostomia. HRUS of left parotid gland demonstrated enlarged parotid gland with heterogeneous parenchyma containing multiple small nodular hypoechoic areas suggesting sialectasis. Few subcentimetric submandibular lymph nodes were also present. Fine needle aspiration cytology (FNAC) of left parotid swelling and enlarged left submandibular lymph node revealed chronic sialadenitis and reactive lymphadenitis respectively.

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DISCUSSION

Juvenile chronic recurrent parotitis is characterized by nonspecific sialadenitis resulting in recurrent episodes of edema of the parotid glands. The first episode typically occurs between the first and second years. In most cases it is not diagnosed; unnoticed or confused with otitis, mumps or pharyngitis. Clinical features of JRP include recurrent parotid inflammation with swelling and pain associated with fever which usually lasts for 2–7 days. This pathology is usually unilateral but can occur bilaterally with symptoms usually more prominent on one side. The natural history of this disease is its recurrence with average number of attacks per year ranging approximately between 1 to 20 (5). The diagnosis is clinical and is usually carried out after the third or fourth episodes due to recurrent history of clinical symptoms (6). The pathogenesis of JRP remains unclear and the present consensus favors a multifactorial origin. Various factors that have been suggested for the development of JRP include congenital ductal malformations, hereditary genetic factors, viral or bacterial infections, allergy, and local manifestation of an autoimmune disease (7,8).

Recent studies conclude that ultrasonography is the investigation of choice for the diagnosis and monitoring of JRP (9). The typical ultrasound shows a thick and edematous parotid gland with heterogeneous echotexture with multiple hypoechoic foci suggesting sialectasis. Multiple hyperechoic central foci of salivary gland secretions may also be seen (10).

This disease shows a predominance in males. The natural history tends to follow spontaneous remission during the pubertal period, but some may persist into adulthood episodes. Recent studies suggest that acute exacerbation of juvenile recurrent parotitis is based on a slowly progressive destruction of the gland (11). The main diagnosis should be differential (12) which may include bacterial epidemic mumps, the parotid duct stones, tumors of the gland and systemic diseases (Mikulick syndrome, Sjögren's syndrome). Viral infections (CMV, EBV, HIV), diabetes mellitus or cystic fibrosis are also included in the differential diagnoses.

The prognosis of JRP is generally good, with spontaneous resolution commonly occurring at adolescence, and so, treatment of the condition should be conservative. Remission of acute attacks may be hastened by the use of sialogogues, gland massage, encouragement of fluid intake, and oral antibiotics (13). More radical treatment is seldom required and is restricted to the severe cases where resolution does not occur. Total parotidectomy is the most effective treatment for adult patients with persistence of JRP and is usually curative, although it carries a risk of facial nerve damage (14). However, it is contraindicated in children, because of the likelihood of spontaneous remission (14).

CONCLUSION

Juvenile recurrent Parotitis is a rare condition whose diagnosis often goes unnoticed. In most cases it undergoes spontaneous remission during childhood but in few adult patients it may persist which may require more radical treatment. High resolution ultrasonographic evaluation of parotid gland, which is a non-invasive and easily affordable investigation, can play a valuable role for timely diagnosis of such a rare condition.

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An Unusual Case of Tendo Achilles Xanthoma

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A xanthoma is a yellowish cholesterol-rich deposition which can appear anywhere in the body representing various diseased state. Tendoachilis xanthoma is the most common tendon xanthoma among them but relatively a rare disease. Ultrasonography might be considered a useful diagnostic and monitoring tool for this infrequent clinical condition. Reported case of a 26 years old male presented with a swelling near right heel developed within a time period of one year associated with occasional pain followed by involvement of left heel which started worsening with walking. High resolution ultrasonography revealed bilateral symmetrical enlargement of achilis tendon with few tiny calcifications and loss of normal fibrillar pattern. Lab investigations reported hyperlipidemia with raised level of serum uric acid and radiological correlation confirmed the diagnosis.

Keywords: Xanthoma, tendoachilis, hypercholesterolemia, hyperuricemia.

Xanthomas are yellowish cholesterol rich deposition or exogenous benign mass, presented on tendons, synovium, and subcutaneous tissues. It can be diagnosed with various imaging in patients with hyperlipidemia and hyperuricemia. Tendon xanthomas are usually diagnostic for heterozygous familial hypercholesterolemia (HFH) and they mostly occur in Achilles tendon. Hyperuricemia combined with xanthoma is very rare. The pathogenesis of hyperuricemia caused by a disorder of purine metabolism and/or a decrease in uric acid excretion, causing deposition of urate crystals in various joints and the surrounding soft tissue with features of joint pain and/or the appearance of gouty nodules, which most often occur at the joints of extremities. We report a case of tendoachilis xanthoma with hypercholesterolemia and hyperuricemia.

CASE REPORT

A 26 years old male presented with the complains of swelling near right heel followed by the left and pain on walking for more than one year. The pain usually exaggerated after over activity and relieved after rest with no obvious nocturnal intensification of pain. He was normotensive and non-diabetic with no history of trauma and no significant family history of such kind of swellings. Within recent two months, the pain in his heels had increased in intensity and not entirely relieved after rest or analgesics. The swelling was increasing too. Physical examination revealed symmetrical thickening and swelling just above the level of heels extending from the lower third of both legs involving the areas of both Achilles tendons. On palpation, the oval shaped swellings appeared firm in consistency and slightly tender on pressing. No muscle wasting or neurovascular deficit was identified. Surrounding skin was smooth and freely mobile. No features of inflammation was noted apparently. (Figure 1).

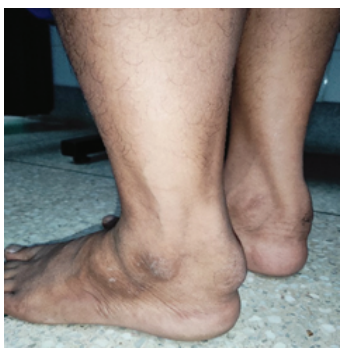

Figure 1: Bilateral symmetrical swellings on Achilles tendons.

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Laboratory investigations of CBC, thyroid function test, urine RME, serum creatinine was within normal reference values. His serum lipid profile shows serum TG (triglyceride) levels of 200 mg/dL, TC (total cholesterol) of 350 mg/dL, LDL (low-density lipoprotein) of 255 mg/dL, HDL (high-density lipoprotein) of 30 mg/dL, and uric acid of 7.6 mg/dL. The normal reference ranges are 50-150 mg/dL, 150-200 mg/dL, <150 mg/dL, >35 mg/dL, and 2-7 mg/dL for TG, TC, LDL, HDL, and uric acid levels respectively.

X-ray revealed no bony abnormalities but showed thickened, noncalcified soft tissue shadows in the region of the Achilles tendon in the both legs (Figure 2).

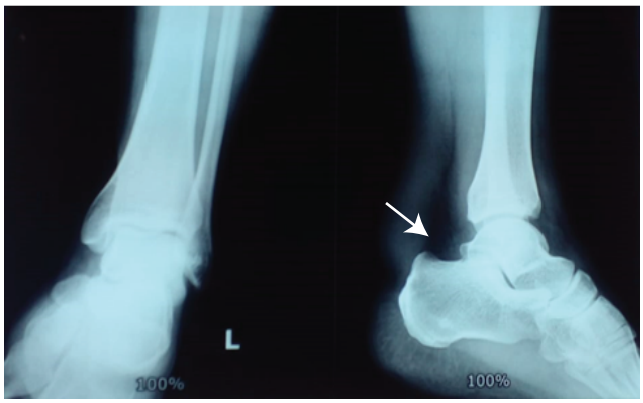
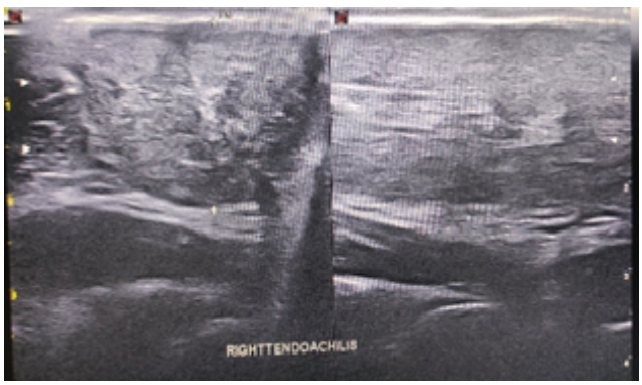


Figure 2: X-ray of left ankle joint with foot. There is enlargement of soft tissue shadows without any calcifications the region of the Achilles tendon. No bony lesion was reported.

Ultrasound scan revealed uniformly thickened hypoechoic Achilles tendon with antero-posterior (AP) thickness of >7 mm, loss of normal fibrillar pattern and multiple tiny calcifications within the tendon (Figure 3).



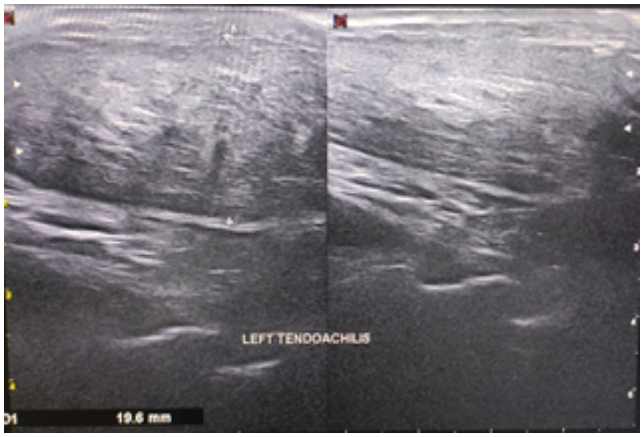


Figure 4: Ultrasound image of thickened Achilles tendon in both a) transverse and b) longitudinal view. Anterior-posterior diameter of right tendon (upper) 20.2 mm and left tendon (lower) 19.6cm with hypoechoic echogenicity. There is also loss of normal fibrillar appearance.

MRI could not be done as the patient was unable to bear the expanses.

The patient is now on lipid lowering agent (atorvastatin), uric acid lowering agent (febustat) and non-steroidal anti-inflammatory drugs. He was also instructed for regular follow up.

Tendoachilis xanthoma is a rare and benign tendon xanthoma (1). It is rarely observed in clinical practice and can easily be confused with neurofibromatosis. In a previous study of a tertiary care hospital in Salem, Tamil Nadu it is found that men have a higher risk of developing Achilles tendon thickening than women (7). Tsouli noted that in most cases, tendinous xanthoma was associated with hyperlipidemia, also in patients with normal blood lipid levels (5). The thickness of the Achilles tendon can therefore be one of the early signs of high cholesterol levels. The clinician can utilize this indicator to evaluate early abnormal cardiac illness. The familial hypercholesterolemia with an autosomal dominant inheritance is characterized by an elevated LDL-cholesterol and tendon xanthomas (2). Low-density lipoprotein (LDL) derived from the circulation accumulates into tendons leading to the transformation of LDL into oxidized LDL (oxLDL)

and the active uptake of oxLDL by macrophages within the tendons which forms xanthomas. Very rarely xanthomas are combined with hyperuricemia or gout (6) . Gouty nodules can occur on any part of joints, including articular cartilage, synovial membranes, joint capsules, tendons, ligaments, or intraosseous or subcutaneous tissue. The joints of the extremities are the most common location (3). A majority of cases are asymptomatic initially and only become symptomatic with increasing size, resulting in weakness of plantar flexion, difficulty in walking, and cosmetic disfigurement, requiring surgery (4). Although Achilles tendon xanthomas (ATX) can be revealed by physical examination there are several imaging methods for their detection. It is worth mentioning that ultrasonography is the method of choice in everyday clinical practice. Sonography is more widely available, inexpensive and an easy technique for the identification of xanthomatosis and is superior to clinical assessment. MRI (Magnetic Resonance imaging) could be also a diagnostic tool but not easily accessible for all patients of our country. Histopathology is the confirmatory tool. Although several treatments for Achilles tendon xanthomas (ATX) have been proposed (LDL apheresis, statins, etc.), they target mostly in the treatment of the basic metabolic disorder of lipid metabolism, which is the main cause of these lesions.

CONCLUSION

Patients with long-term hyperlipidemia, hyperuricemia accompanied by bilateral symmetrical enlargement of the Achilles tendon require early diagnosis to prevent further comorbidity and increase capability. Readily accessible ultrasound imaging helped us ensure maximum patients' benefits in the diagnosis and follow up.

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Bladder Exstrophy: A Report of Three Cases

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Bladder exstrophy is a rare congenital defect of cloacal membrane where the urinary bladder develops outside the fetus resulting in urine leakage. Associated defects in genital, gastrointestinal or musculoskeletal systems may be present. Surgical intervention is the mode of treatment. Few cases have been reported worldwide. Here we have presented three cases of bladder exstrophy in different age & sex. All the cases revealed sonographically non-visualized urinary bladder with anterior abdominal wall defect & deformity in external genitalia. Ultrasonography can play primary role in diagnosing such cases specially in prenatal period.

Keywords: Urinary Bladder, Exstrophy, cloacal membrane, Ultrasonography.

Bladder exstrophy is a rare congenital defect due to abnormality in the development of the cloacal membrane leading to malformation of urinary bladder and urethra in which bladder is turned inside out. Bladder is flattened and exposed to the outside of body through a defect in the lower anterior abdominal wall in midline (1, 2). Symphysis pubis may be absent along with misplaced or deformed external genitalia. In few cases there may be a very small bladder plate (2). Associated intestinal tract as well as musculoskeletal abnormality may be present. It occurs at a rate between 1 in 10,000 to 1 in 50,000 (3) with a male-to-female ratio of 2.3 - 6:1 (4, 5, 6). Familial predisposition has been recorded (5). The only way of management is surgical correction, i.e., bladder, urethral & genital reconstruction along with repair of wall defects.

CASE 1

An 18 days old female child Institute of Nuclear Medicine and Allied Sciences (INMAS), Dhaka with a soft tissue mass protruding from a lower abdominal wall defect, low lying umbilicus and abnormality in the external genitalia (Figure 1). The father complained of continuous dribbling of urine from the urethra when the baby cries. On physical examination the baby had a gap in the lower anterior abdominal wall, in the midline. A pinkish rounded, soft tissue mass of about 2 cm diameter, could be seen through it. Umbilicus was low lying, seen just above the mass and the vulva was placed above its normal position, just below it. The baby had displaced vagina, bifid clitoris, divergent labia and an anteriorly placed anus. She was scanned with a 3.5 and 7 MHz ultrasound probe of a Toshiba nemio-30 USG scanner. The urinary bladder could not be outlined in lower abdomen or anywhere else in the abdominal cavity. Irregular collection was seen in the lower abdomen and between the bowel loops. All other abdominal organs were normal in size with uniform echo pattern. Scan over the mass showed poor passage of echoes with the collection in the lower abdomen. The pubic bones seemed to be wide apart.

The patient underwent bladder reconstruction surgery and came for follow up after 6 months. The visible wall defect in lower abdomen was repaired. A small bladder like structure was noted in lower abdomen containing about 8 ml of fluid (Figure 2). 3 ml fluid could be seen after urination.

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