reperfusion injury following recovery from circulatory shock, over-distended gall bladder (cholestasis) due to 'nil per oral' status, drug- or disease- induced mucosal ulceration of the gut, mesenteric lymphadenitis, associated hepatitis and referred pain from pleurisy [7,10]. Regional lymphadenopathy at the bite site is a characteristic feature of scrub typhus seen in 10-60% of patients [1]. As per the lymphatic anatomy, eschar of the right hip will cause enlargement of ipsilateral iliac nodes. In our patient, this could have contributed to localized tenderness in the right iliac fossa.

Omitting a laparotomy in the presence of actual inflammation would constitute medical negligence; while on the other hand, an unnecessary operation done in the presence of febrile illness is potentially hazardous and avoidable. To resolve this dilemma, several scoring systems have been proposed.

We consider SARS-CoV-2 antibodies noted in our patient to be due to the well documented phenomenon of serological cross-reactivity with dengue [11]. Despite performing laparotomy, we avoided excision of non-inflamed appendix as it may increase the risk of wound infection and add to the morbidity without any additional benefits. This case highlights the need to consider co-infections and rare complications in children with uncommon presentations.

JAN AMRITHA,[1] VENKATACHALAM RAVEENTHIRAN[2]*
From Departments of [1]Pediatrics and [2]Pediatric Surgery, Government Cuddalore Medical College, Chidambaram, Tamil Nadu.
**vrthiran@gmail.com*

REFERENCES

1. Anitharaj V, Gunasekaran D, Pradeep J, Stephen S. Gastrointestinal manifestations of scrub typhus in children and adults from Puducherry and neighbouring Tamil Nadu State, India. J Gastrointest Infect. 2017;7:1-4.
2. Sapkota S, Bhandari S, Sapkota S, Hamal R. Dengue and scrub typhus co-infection in a patient presenting with febrile illness. Case Rep Infect Dis. 2017;2017: 6214083.
3. Iqbal N, Viswanathan S, Remalayam B, et al. Pancreatitis and MODS due to scrub typhus and dengue co-infection. Trop Med Health. 2012;40:19-21.
4. Jayasundara B, Perera L, de Silva A. Dengue fever may mislead the surgeons when it presents as an acute abdomen. Asian Pacific J Trop Med. 2017;10:15-19.
5. Mahajan SK, Babu SN, Sharma D, et al. Scrub typhus presenting as acute abdomen. Trop Doct. 2011;41:185-6.
6. Kumarasena L, Piranavan P, Bandara S, et al. A case of dengue fever with acute appendicitis: not dengue fever mimicking appendicitis. Sri Lanka J Surg. 2014;32: 33-35.
7. Mcfarlane MEC, Plummer JM, Leake PA, et al. Dengue fever mimicking acute appendicitis: A case report. Int J Surg Case Rep. 2013;4:1032-34.
8. Thipmontree W, Suwattanabunpot K, Supputtamonkol Y. Spontaneous splenic rupture caused by scrub typhus. Am J Trop Med Hyg. 2016;95:1284-86.
9. Chang PH, Cheng YP, Chang PS, et al. A case report and literature review of scrub typhus with acute abdomen and septic shock in a child - the role of leukocytoclastic vasculitis and granulysin. Am J Dermatopathol. 2018;40:767-71.
10. Lee CH, Lee JH, Yoon KJ, et al. Peritonitis in patients with scrub typhus. Am J Trop Med Hyg. 2012;86:1046-48.
11. Masyeni S, Santoso MS, Widyaningsih PD, et al. Serological cross-reaction and co-infection of dengue and COVID-19 in Asia: Experience from Indonesia. Int J Infect Dis. 2021;102:152-54.

Recurrent Generalized Scleredema in an Adolescent Girl With Uncontrolled Type 1 Diabetes Mellitus

Scleredema is a non-pitting induration of the skin and connective tissue, usually secondary to intrinsic disease or infection, rare in children, and generally reversible on treating the primary disease [1]. The main associations of scleredema in children are infections [2-4]. Sceleredema as a manifestation of uncontrolled diabetes in children and adolescents is rarely reported in India [5]. We report recurrent generalized scleredema in an adolescent girl having poorly controlled type 1 diabetes mellitus (T1DM).

A 13-year-old girl presented with generalized swelling over the whole body for six months. The swelling started from the arm followed by the abdomen and gradually involved the face, chest and legs. She also complained of pain in the abdomen, arms and legs for one month. There was stiffness of facial muscles and difficulty in opening the mouth and difficulty swallowing food for 15 days. There was no history of cold, cough, fever with rash, morning stiffness in joints, no history of Raynaud phenomenon, or eating raw or undercooked pork. She was diagnosed as T1DM at 9 years of age but was poorly compliant to the prescribed insulin injections. Her documents showed persistent hyperglycemia and frequent hospitalization for diabetic ketoacidosis. She weighed 35 kg, and on examination was alert, afebrile, with heart rate of 90/min, respiratory rate 20/min, blood pressure 108/68mm Hg, and diffuse swelling was seen over the face, thorax, abdomen, bilateral upper and lower limbs. Swelling was diffuse, tense, waxy, hard, non-tender, indurated, non-pitting and could not be pinched. Her face was blank, mask-like, with abolition of skin linings and folds. The rest of the general and systemic examinations were normal.

She was hospitalized and started on insulin as a split-mix regime along with symptomatic treatment. Investigations revealed total leucocyte counts 12.3×10^9/L, hemoglobin12.3 g/dL, alanine aminotransferase 64 U/L, aspartate aminotransferase 28 U/L, total protein 6.6 mg/dL, serum albumin 4.2 g/dL, total cholesterol 192 mg/dL, triglycerides 176 mg/dL, TSH 1.36 µIU/mL, blood urea 26 mg/dL, serum creatinine 0.64 mg/dL, ionized calcium 7.9 mg/dL, serum potassium 4.25 meq/L, sodium 136.2 meq/L . Her random blood glucose was 554 mg/dL, urine glucose

3+and glycated hemoglobin was 13%. Arterial blood gas analysis revealed compensated metabolic acidosis. The HIV serology was negative, rheumatoid factor was 5 IU/mL (normal), anti-streptolysin titer 40 IU/mL and anti-ds DNA titer was negative. Chest radiograph and ultrasonography of abdomen were normal. Skin biopsy from the back of chest was suggestive of sceleredema. The biopsy was stained with alcin blue which revealed normal epidermis, mucin filled thickened collagen bundles with deposition of acid hyaluronidase. Fundoscopy revealed diabetic retinopathy in her both eyes and surgery was recommended.

After one week of achieving normoglycemia, the scleredema started reducing from the abdomen, and upper and lower limbs. She was discharged with scleredema persisting on her face. On follow-up, after two months, her weight reduced to 33 kg and sceleredema resolved completely as nasolabial folds and wrinkles on the forehead reappeared.

After 8 months, she again presented with generalized scleredema all over the body, more on the face as compared to the abdomen and limbs, with poor acceptance of meals. The mother associated worsening of sclererderma with glargine insulin and improvement if glargine insulin was replaced with regular or mixtard insulin. She was admitted and insulin was started as a split-mix regimen. Her blood glucose was high and glycated hemoglobin was 16.8%, liver function tests, renal function tests, erythrocyte sedimentation rate and C-reactive protein were normal. The repeat ASO titer and Anti-ds DNA titer were negative. During recurrence, skin biopsy was not performed due to unavailability of consent from parents. After a few days, blood glucose was controlled, oral intake improved and she was discharged on split-mix regime on mixtard insulin, with advice regarding strict diet and blood glucose control. After two months, on follow up, scleredema was diminished from the abdomen and limbs but persisted on the face. She continues to remain under follow-up subsequently.

Scleredema necessitates morphological classification as a generalized or localized type. Generalized sceleredema can be described as diffuse affection of the entire body, like the face, thorax, abdomen, and upper and lower limbs with less skin hardening. It is common in children irrespective of precipitating etiology [2-5]. Localized scleredema can be explained as a patchy and randomly distributed lesions over either upper or lower half of body or involving non-contiguous multiple sites. Skin thickening and hardness is more in localized type, refractory to treatment, usually seen in obese adults having prolonged poorly controlled type 1 or type 2 diabetes mellitus known as scleredema diabeticorum.

The skin shows hyperplasia of fibroblast and excess collagen synthesis in sclererderma, that can get triggered due to hyperinsulinemia or prolonged hyperglycemia with resistance to insulin [6]. Skin tightening, swelling and hardness found in scleredema mimics scleroderma, scleromyxedema, and trichinella. These were distinguished in this child by confirmation of scleredema on histology, normal TSH, non-ingestion of pork and absence of fibrotic or sclerotic changes in skin. Normal ASO titre and absence of pyoderma, tonsillopharyngitis, common cold, cough, fever with rash demarcated it from infections like streptococcal, influenza, scarlet fever, measles, rubella and mumps.

Non-resolution of scleredema within a year necessitates work up for secondary causes like multiple myeloma, monoclonal gammopathy, paraproteinemias, Sjogren syndrome and hyper-parathyroidism. Normal cardiac, renal and nervous system examination along with normal laboratory reports in the index child excluded these causes. She remained well over the next one year and didn't have any new symptom.

In diabetic obese males older than 40 years of age, scleredema is10 times more prevalent and starts in adulthood [6], which is contrary to the index case who was a 13-year-old slim girl. Chronic and prolonged diabetics on insulin injections having retinopathy are prone to develop scleredema as observed in index child and earlier also [5]. Recurrent episodes are unusual and under-reported, but our patient also experienced a second episode of generalized scleredema associated with glargine insulin. Type of insulin may affect the development of scleredema as is reported earlier [5], where NPH insulin flared scleredema while Lente insulin reduced.

Treatment of scleredema depends on eliminating or controlling the triggering factor [2,5]. A high index of suspicion is needed for diagnosing scleredema in children with chronic diabetes with microvascular complications. The strict control of blood glucose is the mainstay of treatment. It is a rare entity and its reappearance is a rarest possibility.

PRAMILA VERMA,[1*] GARIMA AGRAWAL VARSHNEY,[1] NANDINI DIXIT,[1] SANJAY AGRAWAL[2]

From Departments of [1]Pediatrics and [2]ENT, Nandkumar Singh Chouhan Government Medical College, Khandwa, Madhya Pradesh.

**drpramilav@yahoo.com*

REFERENCES

1. Greenberg LM, Geppert C, Worthen HG, et al. Scleredema adultorum in children. Pediatrics. 1963;32: 1044-54.
2. Gupta K, Gupta R, Mahajan A. Scleredema in a child. JK Science. J Med Educ Res. 2003; 5:26-128.
3. Parmar RC, Bavdekar SB, Bansal SS, et al. Scleredema adultorum. J Postgrad Med. 2000; 46:91-3.
4. Heilbron B, Saxe N. Scleredema in an infant. Arch Dermatol. 1986;122:1417-19.
5. Toyota T , Umezu M, Oikawa N, et al. Diabetic scleredema. Tohoku J Exp Med. 1983;141:457-61.
6. Svoboda SA, Shields BE. Cutaneous manifestations of nutritional excess: Pathophysiologic effects of hyperglycemia and hyperinsulinemia on the skin. Cutis. 2021;107:74-78.