CASE REPORT

Acute pancreatitis with eruptive xanthoma

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SUMMARY

Eruptive xanthomas are benign skin lesions caused by localised deposition of lipids in the dermis. Xanthomas can present as early manifestations of systemic disorders, which are typically caused by elevated levels of serum triglycerides and uncontrolled diabetes. Early recognition and treatment of the underlying condition decreases morbidity and mortality. After treatment of the underlying metabolic disorders, lesions mostly disappear without leaving scars. We present a case with multiple yellowish, erythematous papules on the extremities suggestive of eruptive xanthomas admitted to our hospital with acute pancreatitis.

BACKGROUND

Cutaneous xanthomas develop as a result of intracellular and dermal deposition of lipid. One of the major distinguishing clinical features of xanthomatous tissue is a characteristic yellow to orange hue. Xanthomas may present with a variety of morphologies, from macules and papules to plaques and nodules. Xanthomas can develop in the setting of primary or secondary disorders of lipid metabolism or monoclonal gammopathy. The major forms of xanthomas associated with hyperlipidaemia are eruptive, tuberous, tendinous and plane (including xanthelasma). Hypertriglyceridaemia can lead to complications such as myocardial infarctions, cerebrovascular accidents, hepatic steatosis and acute pancreatitis.¹ Here we report the development of acute pancreatitis due to hypertriglyceridaemia in an eruptive xanthomic case and therapeutic plasma exchange (TPE) administration as a treatment.

Figure 1 Common yellow papules on the right side.

Figure 2 Immunohistochemistry positive staining for histiocytes with CD68 (×200).

CASE PRESENTATION

A 27-year-old male patient was admitted to the dermatology clinic because of the numerous yellowish-brown bumps that lasted for about 6 months on the arms and thighs. There was no pathological finding except the lesions. The patient's body mass index was 31.04 kg/m².

Dermatological examination revealed a large number of yellowish papules ranging from 3 to 6 mm in the arms, elbows and thighs (figure 1). Punch biopsy was obtained. Histopathological examination revealed vacuolar cytoplasmic histiocytes (foam cells) and perivascular lymphocytic infiltration distributed between the collagen fibres in the upper and middle dermis. Immunohistochemically histiocytes stained positively with CD68. The case was found to be compatible with eruptive xanthoma (figure 2). Laboratory findings include triglycerides 105 mmol/L (normal: 0.25–5 mmol/L), total cholesterol 11.5 (normal: 2.8–5 mmol/L), high-density lipoprotein cholesterol 3.8 mmol/L, low-density lipoprotein cholesterol 0.3 mmol/L, aspartate aminotransferase 80 U/L (normal: 5–50 U/L) and alanine transaminase 67 U/L (normal: 5–50 U/L). Fasting blood glucose was 4.3 mmol/L (normal: 1.8–2.5 mmol/L) and HbA1c was 54.1 mmol/mol (normal: 20–42 mmol/mol). Total blood count, thyroid function tests and amylase were normal. On abdominal ultrasound (USG), the liver was in normal size and localisation and the parenchymal echo was seen as compatible with hepatosteatosis (grade 2). Gemfibrozil for hypertriglyceridaemia and metformin for type 2 diabetes mellitus were started. However, after 6 days, the patient was admitted to the emergency department with severe abdominal pain. In laboratory, white blood cell was 21.35 × 10⁹/L (N:

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Figure 3 CT findings consistent with thick hypodense pancreatic tail: acute pancreatitis.

4.5–11 $10^3/\mu\text{L}$), C reactive protein was 179 mg (normal: 0–5 mg), triglyceride was 28.8 mmol/L (normal: 0.25–5 mmol/L) and amylase was 110 U/L (normal: 28–100 U/L). On abdominal CT examination, the pancreas was normal in size and the pancreas tail segment was thick and hypodense, and peripancreatic fatty planes showed dirty infiltrate, minimal local fluid ([figure 3](#)). We were admitted to the endocrinology clinic with clinical, laboratory and radiological confirmation of acute pancreatitis. The patient underwent therapeutic plasma exchange (TPE) twice with fresh frozen plasma and albumin via double membrane filtration. The triglyceride levels were reduced to 20 mmol/L from 80 mmol/L after TPE administration. Remarkably, eruptive

Figure 4 Eruptive xanthoma, completely regressed clinically on the sixth month.

xanthoma lesions completely disappeared at 6 months ([figure 4](#)).

DISCUSSION

Eruptive xanthomas are papules ranging from about 1 to 5 mm in diameter, erythematous or grossly appearing, and usually on the extensor surface of the extremities, in the wrists and hands. As it may be asymptomatic, itching and tenderness may also be seen.¹ In addition, in the eruptive xanthomas, the Koebner phenomenon was seen in some cases in the literature.^{2–4} We have not found any evidence of this phenomenon in our case.

Eruptive xanthomas may be primary or secondary hypertriglyceridaemia. Triglyceride levels in patients with eruptive xanthomas are often over 80 mmol/L and can be seen in hypertriglyceridaemia type 1 (chylomicron increased), type 4 (increased very low density lipoprotein (VLDL)) and type 5 according to the Fredrickson hyperlipidaemia classification.^{1 5} One reason for the elevation of triglyceride levels is the inability to clear circulating lipids. Another cause of elevated triglycerides is liver overproduction of endogenously produced triglyceride-rich lipoproteins. Environmental factors and some diseases cause hypertriglyceridaemia and eruptive xanthoma by affecting triglyceride metabolism worse. Obesity, high calorie intake, uncontrolled diabetes mellitus, hypothyroidism, nephrotic syndrome, alcohol consumption, oral oestrogen replacement and systemic therapies (retinoids, olanzapine, protease inhibitors) and lipoprotein lipase deficiency may cause hypertriglyceridemia.^{1 6 7} There were excess alcohol intake, obesity and diabetes mellitus in our case's history, and we checked lipoprotein lipase deficiency and there was no mutation.

Characteristic histological findings of eruptive xanthomas are foamy macrophages containing lipids in their cytoplasm. In early lesions, the foam cells are small in number and in size. Initially, there is a mix infiltrate rich in neutrophils and lymphocytes. As lipidisation increases, lesion appearance becomes typical xanthoma. Extracellular lipid in the dermis is found.^{1 8 9} In our case, vacuolar cytoplasmic histiocytes (foam cells) and perivascular lymphocytic infiltration distributed between the collagen fibres in the upper and middle dermis were observed, and eruptive xanthoma was confirmed histopathologically.

The purpose of treatment of eruptive xanthoma is to lower the serum triglyceride level and to keep it low. In most cases of eruptive xanthomas, it is not possible to lower the level of excessively high triglyceride to normal levels by drugs immediately. The most common treatment that lowers serum triglyceride levels is fibric acid derivatives and omega-3 fatty acids, but TPE in emergency situations is an effective method for rapid recovery of the patient.¹⁰ The cutaneous lesions due to hypertriglyceridaemia may exist for months.¹¹ Hypertriglyceridaemia

Learning points

- ▶ Xanthomas can develop in the setting of primary or secondary disorders of lipid metabolism or monoclonal gammopathy.
- ▶ Early recognition and treatment of the underlying condition that causes eruptive xanthomas decreases morbidity and mortality.
- ▶ Hypertriglyceridaemia should be kept in mind in patients admitted to the clinic with eruptive lesions, and the diagnosis of eruptive xanthoma must be confirmed pathologically.
- ▶ It can take several months for the lesions to recover.

should be kept in mind in patients admitted to the clinic with eruptive lesions, and the diagnosis of eruptive xanthoma must be confirmed pathologically. It is conceivable that patients with hypertriglyceridaemia may develop acute pancreatitis, and TPE may be considered to achieve rapid lipid clearance in the development of acute pancreatitis. It should also be known that the regression of eruptive xanthomas may take months.

Contributors SD planned the article and wrote the case. ZAS and HSI read and made the changes about the literature. SE examined the pathology specimens.

Competing interests None declared.

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REFERENCES

- 1 Massengale WT, Hodari KT, Boh EE, *et al*. Xanthomas. In: Bologna JL, Jorizzo JL, Schaffer JV, eds. *Dermatology*. 3rd ed. London, England: Mosby Elsevier, 2012:1547–55.
- 2 Goldstein GD. The Koebner response with eruptive xanthomas. *J Am Acad Dermatol* 1984;10:1064–5.
- 3 Miwa N, Kanzaki T. The Koebner phenomenon in eruptive xanthoma. *J Dermatol* 1992;19:48–50.
- 4 Miwa N, Kanzaki T. The koebner phenomenon in eruptive xanthoma. *J Dermatol* 1992;19:48–50.
- 5 Pai VV, Shukla P, Bhobe M. Combined planar and eruptive xanthoma in a patient with type IIa hyperlipoproteinemia. *Indian J Dermatol Venereol Leprol* 2014;80:467–9.
- 6 Park JR, Jung TS, Jung JH, *et al*. A case of hypothyroidism and type 2 diabetes associated with type V hyperlipoproteinemia and eruptive xanthomas. *J Korean Med Sci* 2005;20:502–5.
- 7 Chang JH, Lee SH, Lee SJ, *et al*. A case of type V hyperlipoproteinemia and xanthoma eruptivum associated with diabetes mellitus. *Korean J Dermatol* 2000;38:561–3.
- 8 Bickley L. Yellow papules on a middle-aged woman. Eruptive xanthomatosis (EX). *Arch Dermatol* 1989;125:289:288–289.
- 9 Crowe MJ, Gross DJ. Eruptive xanthoma. *Cutis* 1992;50:31–2.
- 10 Rader DJ, Hobbs HH. Disorders in Lipoprotein Metabolism. In: Kasper DL, Fauci AS, Longo DL, *Harrison's Principles of Internal Medicine*. 18th ed. New York: NY: McGraw-Hill, 2012:3145.
- 11 Bologna J, Jorizzo JL, Schaffer J. Xanthomas. In: *Dermatology*. 2nd ed. Philadelphia PA: Elsevier Saunders, 2012:1411–9.

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