Letters

AN UNUSUAL CASE OF LOCALISED HYPERTRICHOSIS

Editor

Topical testosterone is now a widely used mode of testosterone replacement therapy. It is well reported that transcutaneous absorption of testosterone may lead to hirsuitism and virilisation.\(^1^,\)\(^2\) We report an interesting case of localised hypertrichosis on the forearm of a female patient and postulate that this was the result of accidental transfer of testosterone gel from the patients’ husband.

A 66-year old lady presented with a one-year history of localised hair growth on the right forearm. She denied excess hair growth or alopecia elsewhere and did not report any other signs of virilisation. The patient had no other relevant past medical or drug history. Closer questioning revealed that she had been applying a 5% testosterone gel (testogel\textsuperscript{®}) with her right hand to her husbands shoulder, intermittently for 4 years. This was applied daily for hypoandrogenism, secondary to radiation therapy for multiple myeloma.

On examination she had localised hypertrichosis on the right forearm, sparing the right dorsal hand, associated with an eczematous eruption. (Fig. 1) There were no other relevant clinical findings. Hormone profile including free testosterone was normal. In view of the temporal relationship between application of the gel and the findings of localised hypertrichosis on the forearm of a female patient, we feel that the intermittent application of testogel\textsuperscript{®} was the causative factor.

In relation to topical testosterone, there have been recent case reports of precocious puberty in children and hirsuitism\textsuperscript{+/-} virilisation in women following accidental transfer of topical testosterone.\(^3^\) One recent case report and review of the literature also described progressive hirsuitism in a premenopausal woman associated with fluctuating testosterone levels of 1.6-6.7 over a 3-month period (normal range<2.5).\(^3\) This was felt to be secondary to transfer of testosterone gel from her partner during contact, because her hair growth and testosterone levels returned to normal after her partner switched to injectable testosterone.\(^3^\) Not all cases are associated with hyperandrogenism however. In one case series of two females applying testosterone gel for treatment of lichen sclerosus et atrophicus, both developed hirsuitism two months later.\(^1\) Hormonal profiles were normal in both cases, however, the gel had been discontinued several weeks before presentation.\(^1\)

Localised hypertrichosis is another known side effect of topical testosterone gel. In a recently published study looking at the effect of transdermal testosterone in female patients, the investigators found that the most common side effect was dose-related hypertrichosis, predominantly at the delivery site.\(^3\) To our knowledge however, there are no reported cases of localised hypertrichosis secondary to inadvertent transfer of topical testosterone between two people.

Regarding the onset of hirsuitism and virilisation with testosterone, time to development varies between reports, most cases presenting between 8-72 months of use and resolving within 2-12 months.\(^1\)

This interesting case highlights the importance of a thorough history in a patient presenting with hirsuitism, hypertrichosis or virilisation, particularly when the pattern of hair growth is unusual in the presence of normal hormonal investigations.

The authors have no conflict of interest

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REFERENCES

1. Hernandez-Nunez A, Dauden E, Garcia-F-Villata MJ, Rios-Buceta L, Garcia-Diez A. Hirsuitism secondary to topical testosterone: report of two cases and review of the literature. J Eur Acad Dermatol Venereol. 2004;18(2):208-210.
2. De Ronde W. Hyperandrogenism after transfer of topical testosterone gel: case report and review of published and unpublished studies. Hum Reprod. 2009;24(2):425-28.
3. Davis S, Papilia MA, Norman RJ, O’Neill S, Redelman M, Williamson M, et al. Safety and efficacy of a testosterone metered-dose transdermal spray for treating decreased sexual satisfaction in premenopausal women: a randomized trial. Ann Intern Med. 2008;148:569-577.

PANCREATIC HETEROTOPIA PRESENTING AS A GASTRIC SUBMUCOSAL LESION

Editor

Heterotopia is the normal tissue of an organ found at an abnormal site without anatomic and vascular continuity from the original organ. It is thought that this arises during embryonic development, where groups of cells differentiate in a manner which is inappropriate for their anatomical position in the body.\(^1^,\)\(^2\) The usual gastrointestinal sites of Pancreatic Heterotopia (PH) include stomach, duodenum, jejunum, Meckels diverticulum, and gallbladder.\(^3\) The condition is
relatively infrequent and usually asymptomatic with post mortem prevalences ranging from 0.6% to 13.7%.

Rare symptomatic cases do arise causing dyspepsia, abdominal pain, melaena, anaemia, nausea and obstruction.

We report a case of PH presenting as intermittent gastric outlet obstruction. A 43 year old man presented with a 4 month history of intermittent post prandial epigastric pain and nausea. Complete gastric obstruction was not evident.

An upper gastrointestinal endoscopy revealed a 3cm lesion at the pylorus. (Fig 1) Ultrasound did not highlight any other cause for upper abdominal pain.

Endoscopic ultrasound (EUS) of the lesion confirmed it to be situated within the submucosa having morphological characteristics suggestive of a gastro intestinal stromal tumour (GIST). (Fig 2) EUS can demonstrate echogenic differences between different types of submucosal lesions and the depth of its invasion. Characteristic EUS features highly suggestive of PH tissue are hypoechogenicity or heteroechogenic structure. Anechoic areas usually correlate with ductal structures. These commonly arise from the third or fourth EUS layers of the GI tract or a combination of both.

GIST also originate from the fourth layer of the GI tract and the presence of cystic spaces can indicate a risk of malignant change. The difficulty in diagnosis requires histological confirmation for a definitive answer.

CT scan confirmed the endoscopic ultrasound findings. There was no evidence of distant metastatic spread. (Fig 3) Retrospective study of CT appearances of gastric submucosal lesions shows that by using a list of specific CT criteria PH can be differentiated from small gastro intestinal stromal tumours or leiomyoma with a high degree of accuracy.

Subsequently the patient proceeded to laparotomy where a 3cm lesion was located in the pyloric channel. A distal gastrectomy was undertaken and the patient made an uneventful recovery. Review in the outpatient department several months following surgery confirmed the relief of his symptoms. Histology revealed the lesion to be consistent with a focus of PH encompassing a cystically dilated duct. (Fig 4)
COMMENT

PH is part of the differential diagnosis of gastric submucosal nodules. The likely aetiology of PH is congenital and usually asymptomatic. However if symptoms occur they are usually in the fourth and fifth decades. PH is a rare differential diagnosis of a submucosal gastric lesion.

The distribution of PH is 25% in the stomach and 30% in the duodenum with the rest distributed at other sites throughout the gastrointestinal tract. There is also the exceedingly rare possibility of malignant change.

This case highlights the rare aetiology of a symptomatic gastric submucosal lesion as well as the difficulty in making a preoperative diagnosis even with modern imaging modalities such as CT and EUS.

The authors have no conflict of interest

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REFERENCES

1. Lackie JM, Mor IAR. Cells and tissues in health and disease. Liver, biliary tract and pancreas. In: MacSween RNM, Whaley K, editors. Muir’s Textbook of pathology. 13th ed. London: Edward Arnold; 1992. p. 33798
2. Mowat A, MacSween RNM, Percy-Robb IW, Foulis AK. Liver, biliary tract and pancreas. In: MacSween RNM, Whaley K, editors. Muir’s Textbook of pathology. 13th ed. London: Edward Arnold; 1992. p. 798
3. Bhana BD, Chetty R. Heterotopic pancreas – an unusual cause of cholecytitis. S Afr J Surg. 1999;37(4):105-7
4. Dolan RV, ReMine WH, Dockerty MB. The fate of heterotopic pancreatic tissue: a study of 212 cases. Arch Surg. 1974;109(6):762-5
5. Tonkin RD, Field TE, Wykes PR. Pancreatic heterotopia as a cause of dyspepsia. Gut 1962;3:135-9
6. Chetty R, Weinreb I. Gastric neuroendocrine carcinoma arising from heterotopic pancreatic tissue. J Clin Pathol 2004;57(3):314-7
7. Zinkiewicz K, Juszkiewicz W, Zgodański W, Szumilo J, Cwik G, Furtak J, et al. Ectopic pancreas: endoscopic, ultrasound and radiological features. Folia Morphol (Warsz) 2003;62(3):205-9.
8. Kim JY, Lee JM, Kim KW, Park HS, Choi JY, Kim SH, et al. Ectopic pancreas: CT findings with emphasis on differentiation from small gastrointestinal stromal tumor and leiomyoma. Radiology. 2009;252(1):92-100.
9. Pearson S. Aberrant pancreas. Review of the literature and report of three cases, one of which produced common and pancreatic duct obstruction. AMA Arch Surg. 1951;63(2):168-86.
10. Denève E, Ramos J, Auffort S, Marchand JP, Roussel T, Perrochia H, et al. [Endocrine tumor arising in heterotopic gastric pancreas.] French. Gastroenterol Clin Biol. 2008 Feb;32(2):195-201.
11. Chetty R, Weinreb I. Gastric neuroendocrine carcinoma arising from heterotopic pancreatic tissue. J Clin Pathol 2004;57(3):314-7

POST-OPERATIVE PYODERMA GANGRENOSUM IN ASSOCIATION WITH ILEAL CARCINOID TUMOUR

Editor

Pyoderma gangrenosum (PG) is an uncommon, progressive ulcerative condition of skin. It presents with deep ulceration characterised by an overhanging violaceous border, which can occur on any body surface. It is frequently confused with other more common ulcerating skin conditions such as necrotising fasciitis, vasculitis, purulent drug reactions and skin infections. Since surgery may be used to treat some of these conditions, but is relatively contraindicated in PG, early diagnosis is critical and is usually made in conjunction with a dermatologist.

This 76-year-old male had a laparoscopic assisted right hemi-colectomy for an apparent ascending colonic tumour, however histology actually revealed a well differentiated neuroendocrine tumour of the terminal ileum. Serum pancreatic polypeptide, N and C-terminal glucagon, chromogranin A and urinary 5-HIAA collection were all elevated.

On day 7 this man’s left iliac fossa port site was noted to be indurated and erythematous. Cefuroxime was empirically commenced for a presumed wound infection. He became pyrexic with a leukocytosis of 30,000 mm³ and skin at the port site quickly became sloughy and ischaemic (Figure 1). Following debridement he required transfer to intensive care as a case of suspected necrotising fasciitis.

The patient’s necrotising skin condition progressed relentlessly. He required 4 further debridements with intermittent returns to the intensive care unit for supportive therapy (Figure 2). Microbiology of the skin specimens was insignificant and pathology described neutrophilic abscesses with no evidence of vasculitis, granulomatous inflammation or metastatic tumour. Following a dermatological opinion a diagnosis of PG was made.

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