

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 hirsutism 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®) with her right hand to her husband's shoulder, intermittently for 4 years. This was applied daily for hypoandrogenism, secondary to radiation therapy for multiple myeloma.



Fig 1.

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 with application of the gel and the findings of localised hypertrichosis with a normal hormone profile, we feel that the intermittent application of testogel® was the causative factor.

In relation to topical testosterone, there have been recent case reports of precocious puberty in children and hirsutism +/- virilisation in women following accidental transfer of topical testosterone.² One recent case report and review of the literature also described progressive hirsutism in a premenopausal woman associated with fluctuating testosterone levels of 1.6-6.7 over a 3-month period (normal range <2.5).² 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.² 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 hirsutism 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 commonest 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 hirsutism 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 hirsutism, 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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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, Meckel's diverticulum, and gallbladder³. The condition is



Fig 1. Gastroscopy view

relatively infrequent and usually asymptomatic with post-mortem prevalences ranging from 0.6% to 13.7%⁴.

Rare symptomatic cases do arise causing dyspepsia, abdominal pain, melaena, anaemia, nausea and obstruction^{5,6}.

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.



Fig 2. Endoscopic ultrasound of gastric submucosal lesion

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 structure⁷. 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.



Fig 3. CT scan axial view. Red arrow: gastric submucosal lesion

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 accuracy⁸.

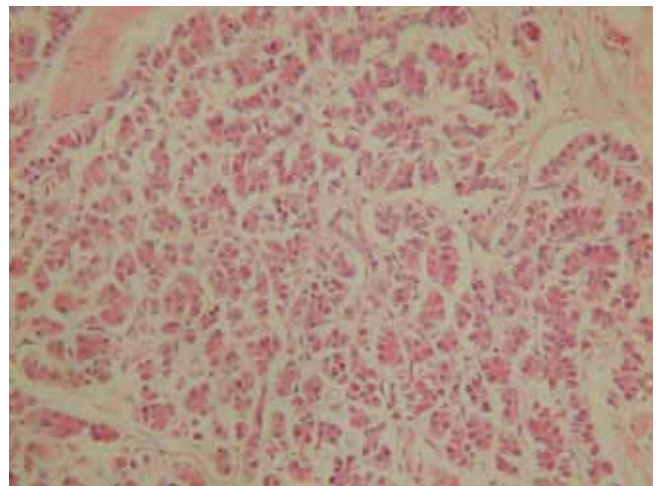


Fig 4. Pancreatic exocrine glands

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 decades⁵. PH is a rare differential diagnosis of a submucosal gastric lesion.

The distribution of PH is 25% in the stomach and 30% in the duodenum⁹ with the rest distributed at other sites throughout the gastrointestinal tract. There is also the exceedingly rare possibility of malignant change^{10,11}.

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.

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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, pustular 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.



Fig 1. Early violaceous change around a laparoscopic port site

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 pyrexial 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.



Fig 2. Extensive abdominal wall debridement with classical violaceous borders seen at the wound periphery

Intravenous antibiotics were stopped and high dose prednisolone was commenced in addition to the already prescribed somatostatin (Octreotide®). The patient was maintained on azathioprine (Imuran®) once the prednisolone had been tapered. His large abdominal defect was dressed with Activon tulle® honey dressings. He progressed well and was discharged. Follow up revealed satisfactory recovery of the wound.

DISCUSSION

The literature yields only one other case connecting PG with carcinoid tumour¹, while most reports correlate the occurrence of PG to trauma, typically surgery². The delay in the recognition of this serious dermatological condition was associated with increased morbidity for our patient. PG is a serious and potentially fatal skin condition when correct treatment is not quickly commenced. Management is relatively simple once recognised with the use of corticosteroids and immunosuppressant. Surgery is not thought to be beneficial and in many circumstances can worsen the condition³.

We recommend that in any significant skin condition, particularly post-operatively or in one not responding to treatment effectively, one must seek the early advice of a dermatologist and not be guided primarily by histology.

The authors have no conflict of interest.

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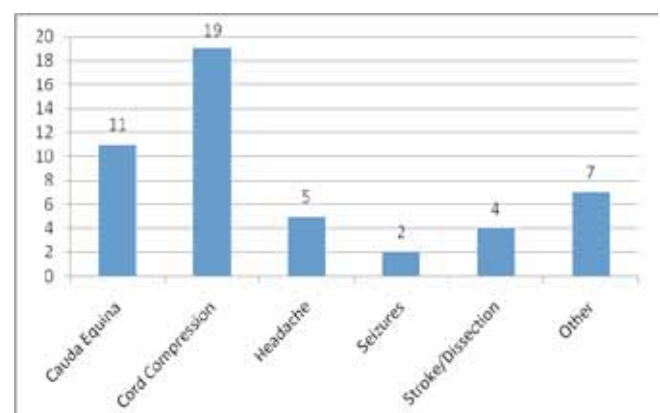
USE OF OUT OF HOURS MRI IN THE ROYAL VICTORIA HOSPITAL – A 6 MONTH RETROSPECTIVE REVIEW

Editor

Through the ongoing development of the Critical Care Centre, it is anticipated that the region's principal trauma receiving unit at the Royal Victoria Hospital will attain Level 1 Trauma Centre status. However an essential criterion for this is the provision of 24 hour access to MRI, as stipulated by the American College of Critical Care Medicine¹. Out of hours MRI is currently provided as a time-limited, daily service on a consultant to consultant referral basis. Within the UK, it has been reported that only 32 out of 88 (36.3%) trauma units with MRI provide an out of hours service².

We undertook a 6 month retrospective review of all patients requiring out of hours MRI between November 2007 and May 2008. Records were assessed for referral information, imaging result and clinical outcome. 74 patients in total had out of hours MRI. Of these, 48 were regarded as emergency (scan performed <24 hours from referral).

Of the 48 emergency requests, the majority came from neurosurgery (n=27) and neurology (n=14), with orthopaedics (n=5), general medicine (n=1) and A&E (n=1) making up the remainder. Figure 1 illustrates the categories of clinical referral, with the majority for either suspected cauda equina syndrome or cord compression.



Out of hours MRI had the greatest impact in suspected cauda equina syndrome, as all scan positive patients (n=5) had surgery on the day of scanning, and made good neurological recovery, with only 1 having ongoing pain at 6 month follow-

up. Early surgery (<24hours) is felt to be of most benefit to those presenting with incomplete cauda equina syndrome³. However, it should be noted that suspected cauda equina syndrome contributed to 15% (11/74) of the total out of hours MRI caseload.

Of the 19 patients investigated for cord compression, 7 were confirmed on MRI. A further 2 patients were diagnosed with cord ischaemia. The remainder were either normal, had degenerative change or disc protrusion not causing compromise of the cord or nerve roots. 2 patients with confirmed cord compression were treated conservatively. Of those who had decompressive surgery, 2 were operated upon within 24 hours of their scan but neurological deficit persisted upon discharge.

It is anticipated that a modern, safe and comprehensive out of hours MRI service to Northern Ireland could be achieved with the 4 district general hospitals which have MRI capacity adopting an out of hours service similar to the current at the Royal Victoria Hospital, coupled with expansion of the Royal Victoria Hospital service to provide 24 hour access. Demand for out of hours MRI is anticipated to further increase with full implementation of NICE guidelines for stroke imaging and suspected metastatic cord compression.

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OLANZAPINE INDUCED HYPONATRAEMIA

Editor

We report a case, a 48 years old woman, presenting with life threatening severe hyponatraemia caused by the Syndrome of Inappropriate Secretion of Antidiuretic Hormone (SIADH) secondary to Olanzapine use. A Medline search revealed no publications of Olanzapine induced SIADH or hyponatraemia. However, online, there were three cases with hyponatraemia been reported at a Dutch pharmacovigilance centre¹.

A 48 years old Caucasian female, obese (BMI 32), smoker with medical history of mixed bipolar affective disorder, schizoid personality disorder and hypercholesterolaemia was admitted to the hospital in a postictal confusional state

following an episode of generalised tonic clonic seizure at home with biting of the tongue and urinary incontinence. There was one day history of generalised muscle aches, anorexia, lethargy, irritability, confusion and unsteady gait prior to the episode. There was no history of polydipsia or polyuria. Shortly after admission, she had respiratory arrest for which she was intubated, started on mechanical ventilation and transferred to ICU.

She was on Olanzapine 20 mg daily for last two years. Her concomitant medications included Diazepam 5mg and Simvastatin 40 mg per day. She had not used any other medication known to cause SIADH during the previous two years. Laboratory investigations revealed hyponatraemia with sodium value of 114 mmol/l, serum osmolality 240 mos/kg, urinary sodium 49 mmol/l and urinary osmolality 220 mos/kg.

Diagnosis of SIADH was made. Olanzapine was incriminated as the causative agent since no other apparent cause of SIADH was found. With discontinuation of Olanzapine and treatment with hypertonic/ normal saline, her serum sodium levels normalised, her respiratory functions improved dramatically and soon, she was weaned off the ventilator, extubated and sent to general ward. In the ward, she continued to maintain normal sodium levels with the discontinuation of Olanzapine. Causality assessment using the Naranjo Nomogram revealed a probable association, with probability score of six.

DISCUSSION

Hyponatraemia (serum sodium concentration < 136 mEq/L) is a prevalent and potentially dangerous medical comorbidity in psychiatric patients². Hyponatraemia is known to occur as a rare but clinically important adverse reaction to treatment with different psychotropic drugs³. In these patients, it is important to rule out psychogenic polydipsia, a clinical disorder characterised by polyuria and polydipsia, as it occurs in 6% to 20% of psychiatric patients and is more likely to be seen in schizophrenia⁴.

In our patient, diagnosis of hyponatraemia secondary to SIADH was made as the biochemical blood and urine test results were consistent with SIADH. SIADH is suspected in any patient with hyponatraemia, hypoosmolality, and a urine osmolality >100 mOsm/kg. It causes hyponatraemia by preventing the excretion of ingested water⁵.

Usually, rapid and complete recovery of drug-induced SIADH occurs when the offending agent is discontinued. In our patient also, the correction of hyponatraemia, combined with the discontinuation of her Olanzapine, resulted in resolution of hyponatraemia, without any further recurrence.

CONCLUSION

Clinicians should be aware that patients being treated with Olanzapine can develop hyponatraemia and it is important to check serum sodium levels when patients on Olanzapine develop symptoms suggestive of hyponatraemia.

The authors have no conflict of interest

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