CONGENITAL RECTO-CUTANEOUS FISTULA

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Abstract
Congenital recto-cutaneous fistula is an uncommon finding in the paediatric population. The pathophysiology of these fistulas is not entirely understood. We present an interesting case of congenital recto-cutaneous fistula. A newborn female was found to have a pit at the inferior gluteal-fold. MRI defined a tract leading to the rectum. At six months and two years of age the fistula became symptomatic and on both occasions she recovered with conservative treatment. Repeat MRI at two years of age demonstrated the fistula tract shortened. At 5 years old the child required drainage of a buttock abscess. Examination could not demonstrate an internal fistula opening. A third MRI demonstrated a persistent tract, which was excised under ultrasound guidance. Histopathology reported a squamous epithelial-lined tract surrounded by chronic inflammation. Congenital recto-cutaneous fistula is an interesting yet not fully understood pathology and we present a case that adds to the complexity of the anomaly.

Keywords: congenital recto-cutaneous fistula, fistula tract

Introduction
Recto-cutaneous fistula is not an unknown finding to Paediatric Surgeons in conjunction with an anorectal malformation, a complication of previous perianal surgery, or older patients with inflammatory bowel conditions. A recto-cutaneous fistula present at birth with a normal anus is an unusual pathology. There have been no cases reported in literature. We discuss the child's presentation, imaging, management, and outcome.

Case Presentation
A newborn girl was found to have a pit at the right-sided inferior gluteal-fold. She was well with no pertinent antenatal history. An MRI soon after birth, due to suspicion of spina bifida occulta, demonstrated a defined tract from the inferior gluteal-fold pit to the rectum. The fistula coursed deep to the subcutaneous fat and overlaid the fascia of the adductor magnus and semitendinosus muscles and passed posterior to the right ischial tuberosity and a second segment extending laterally deep to the muscle and adherent to the ischium (Fig. 1). The fistula became symptomatic at six months and the tract was treated conservatively. A fistulogram under ultrasound guidance failed on two occasions. At two years she developed right buttock inflammation and large purulent discharge from the fistula. Repeat MRI demonstrated the fistula tract shortened to <6 centimetres in length and was managed conservatively. At 5 years the child required drainage of a right buttock abscess. At operation, examination could not demonstrate an internal fistula opening. A third MRI 6 weeks post-surgery demonstrated a 3cm tract from the pit located at the right infra-gluteal crease (Fig. 2). The tract was now also visible on ultrasound and amenable to surgery without gross dissection and tissue damage (Fig. 3).

At surgery under ultrasound guidance, a branching tract was found extending from the opening at the infra-gluteal crease with one segment leading deep to the area of the ischial tuberosity and a second segment extending laterally deep to the muscle and adherent to the ischium. Histopathology reported a squamous epithelial-lined tract surrounded by chronic inflammation. She is asymptomatic at 2
Figure 1. Presenting MRI image with arrow demonstrating fistula tract

Figure 2. Repeat MRI image at 5 years old with arrow demonstrating fistula tract

Figure 3. Ultrasound image of fistula tract at 5 years old

Discussion
Congenital recto-cutaneous fistula is generally associated with congenital anorectal malformations or these fistulas can be the result from previous surgery to correct these malformations. There is also an abundance of published cases of recto-cutaneous fistulas occurring in adults suffering from bowel related pathology such as Crohn’s disease and ulcerative colitis.

A literature search fails to find any reported cases of children born with a congenital recto-cutaneous fistula in the presence of a normal anus and rectum. There is one case reported more than 60 years ago that describes an anal fistula that opened onto the thigh in a 37 year old gentleman with chronic ulcerative colitis [1]. A similar appearing pathology would be the fistula-in-ano. The aetiology of these are not completely understood but hypotheses include abnormal anal glands that become infected, infected anal fissures, excess androgens, entrapment of migratory cells in the hind gut, and abnormal crypts of Morgagni.

As demonstrated in this case, MRI was able to define the fistula tract. MRI has been shown to be superior to CT and fistulograms for the imaging of fistula tracts and for demonstrating the perianal anatomy. The superiority of MRI to CT is the lack of radiation dose and the higher quality resolution of soft tissue planes. Although a fistulogram may demonstrate the path of the tract, it would give limited information of the anatomic relation of the tract to its surroundings and the procedure itself can be painful and carries the risk of introducing infection and causing false passages [2].

This case is interesting from the point of view that this is a congenital fistula tract that had an incidental finding and thus the natural history could be followed through serial infectious presentations and imaging. Also, the fistula tract had a relatively long route that extended from the rectal area to the inferior gluteal crease. There have been no previous reports of fistula tracts opening in this area in children in literature and there is no obvious embryological reasoning for the fistula to follow this particular course.

Conclusion
Congenital recto-cutaneous fistula is an interesting pathology that has not been previously described in literature. This anomaly is complex and its diagnosis and management can be challenging, however treatment of this entity is curative.
REFERENCES
