Case-based review

Subcutaneous fistula in infancy: a different diagnosis of a perianal discharging nodule

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Summary

Fistula-in-ano (FIA) is an abnormal communication between the anal canal and the perianal skin with an external exit hole draining spontaneously. In FIA the fistulous tract starts from a deep acute purulent inflammation of the anal region, usually an abscess. Perianal abscess (PA) generally arises from the acute infection of a small gland just inside the anus due to the entry of bacteria or foreign material in the gland. Fistula-in-ano (FIA) or anorectal fistula is an abnormal communication between the anal canal and the perianal skin with an external exit hole draining spontaneously. In FIA the fistulous tract starts from a deep acute purulent inflammation of the anal region, usually an abscess.

Introduction

Perianal abscess (PA) is a cavity filled with pus located near to the anus or rectum. PA usually arises from the acute infection of a small gland just inside the anus due to the entry of bacteria or foreign material in the gland. Fistula-in-ano (FIA) or anorectal fistula is an abnormal communication between the anal canal and the perianal skin with an external exit hole draining spontaneously. In FIA the fistulous tract starts from a deep acute purulent inflammation of the anal region, usually an abscess.

To date, despite many reports have been published in medical literature, FIA and PA in children still represent a perplexing topic for paediatricians, paediatric surgeons and dermatologists, in fact the boundaries between the two forms are frequently poorly defined. Piazza et al. have reported that PA is normally the forerunner sign of FIA while it is exceptional to find FIA alone (1). In fact most reports of FIA start with PA presenting as a red-violet swelling area.

The Parks classification system of FIA is still commonly used in order to define this condition (2). Unlike the current procedural terminology coding, the Parks classification system does not include the superficial subcutaneous fistula. These fistulae are not of cryptoglandular origin but are usually caused by unhealed anal fissures or anorectal procedures, such as hemorrhoidectomy or sphincterectomy.

Finally, in children PA and FIA appear seldom related to underlying conditions like congenital malformations, immunodeficiency, inflammatory bowel diseases, Hirschprung disease, actinomycosis, tuberculosis, chlamydial infection or iatrogenic trauma (3).

We report a case of FIA unprecedented by PA in an infant.

Case report

A 8-month-old boy was referred to our Department for evaluation of a perianal lesion firstly noticed 3 months before consultation. The baby, born at term, after an uneventful pregnancy, with a birth weight of 3.950 kg, had personal and family history unremarkable.

Physical examination revealed a solitary, little, red, swelling, dome-shaped nodule measuring 0.6 cm in diameter, located in the perianal region on the right side at 2 o’clock position. In the center of the nodule there was a point sized opening and throughout it the mother reported the occasional discharge of a colourless and odourless liquid (Figure 1). The baby ap-
peared otherwise healthy and he was completely asymptomatic even after palpation. There was no involvement of regional lymph nodes and haematological and biochemical routine tests were within the normal values. In particular leukocyte count, erythrocyte sedimentation rate (ESR) and C reactive protein were not contributory. No other cutaneous or extracutaneous abnormalities could be detected. A diagnosis of fistula-in-ano (FIA) was clinically suspected and our patient was referred to the Department of Paediatric Surgery for a surgical evaluation.

Diagnosis of FIA was confirmed in the operative room: in general anaesthesia, the nodular lesion was drained and a silver probe was inserted in the fistulous path starting from the pectinate line. A non absorbable suture was then positioned through the fistula (Figure 2) in order to obtain a healing by second intention thanks to frequent scarifications and rejections of the suture (Figures 3, 4). The first surgery procedure was unsuccessful, so, after 30 days, a fistulotomy, surgical opening and simple division of the tract, was made and the baby healed completely. In order to avoid anal incontinence fistulectomy, surgical excision of the complete tract of the fistula, was not performed. Oral antibiotic therapy was postponed till healing. Follow up at 3 and 6 months did not reveal recurrence. After one year follow-up, our patient is in good health without relapses.

Discussion

We describe a 8-month-old boy, otherwise healthy, with a FIA presenting as a little, red, discharging nodule situated in perianal area, developed at the age of 5 months. This condition has been rarely described in literature. In our case the mother confirmed that the disease was firstly noted as a discharging little nodule in her child.

This clinical presentation seems peculiar, because of the absence of a previous PA, but maybe it should be more common in the first year of life than we suppose. Barthès-Anidjar et al. have reported that in the first year of life PA and FIA are associated and, in contrast
to other age groups, in infants they seem to have mainly a malformative origin, without the presence of an underlying concomitant disease (4). The cryptoglandular hypothesis states that an infection begins in an anal gland and progresses into the muscular wall of the anal sphincters to cause an anorectal abscess and, subsequently, a fistula (5-7). In our case a superficial subcutaneous fistula was detected but no anorectal procedures had preceded the FIA formation. Given that anorectal procedures are rarely performed in infancy, fissures seem more likely to be related to subcutaneous FIA etiology. According to us this kind of fistula should be quite common in infants who did not develop an anticipating abscess, supporting their relation to fissures which are, on the contrary, commonly found in small children. We hypothesize that a superficial infection, generated from an anal asymptomatic fissure, could have caused in our patient this singular condition in which, from the beginning, a little discharging nodule was seen close to the anus, without the presence of a former abscess. During the surgical act the finding of a very short fistula and of a superficial discharging nodule seem to support our diagnosis of subcutaneous fistula.

This case report suggests that subcutaneous fistulae should be considered by pediatricians and dermatologists in the differential diagnosis of all perianal nodules, especially in patients aged less than one year. At any rate, a surgical evaluation has always to be requested in order to confirm the diagnosis and to identify FIA and PA correct management (8). PA in infants is commonly complicated by FIA (3) and the rate of this progression ranges from 20 to 80% (5) suggesting that they may represent different clinical manifestations resulting from the same pathogenetic process (9).

Recent studies have suggested that this evolution is influenced from the treatment approach: conservative management of PA with local care (sulphur baths, local hygiene) and systemic antibiotics, is associated with a rate of FIA formation lower than surgical incision and drainage (5).
In literature there are controversies regarding PA and FIA management, mainly in the cases, like ours, where PA and FIA are not associated to fever and systemic signs of sepsis, because a standardized treatment is not available. Serour et al. (7) documented the spontaneous closure of FIA in at least one-third of their cases, so they proposed a conservative approach consisting only in 1-3 months follow-up. For persisting FIA the fistulotomy with cryptotomy is recommended and laying open the wound is the preferred treatment. In our case surgeons treated the inflamed crypt placing a seton in order to drain the pus without cryptotomy and the patient had no recurrences up to now (10). In order to avoid recurrence it seems to be important the identification and treatment of the abnormal inflamed crypt, as shown in the literature by several studies (3, 4). Furthermore fistulotomy, in this group of age, is safe because FIA in infants is situated superficially without involving anal sphincters and this make the risk of muscle injury remote, in fact we have never documented this complication.

References