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Case Report

A rare case of polythelia on the buttock, found incidentally during colonoscopy

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ABSTRACT

Polythelia or supernummary nipples is a congenital anomaly where more than two nipple- areolar complexes are present. While polythelia is the most common mammary tissue malformation, it is still a rare condition with an incidence of 0.2% to 5.6%. Suppernummary nipples are usually located in the medial embryonic milk lines however, there have been some published cases of atypical locations. We present a case of a 42-year-old male who was incidentally found to have a supernummary nipple on his left buttock on colonoscopy. There was no prior family history of polythelia or any congenital abnormalities.

Keywords: Polythelia, Supernumerary nipple, Buttock

INTRODUCTION

Polythelia, also known as a supernumerary nipple, is a congenital anomaly and presents as a brown-pink macule which is usually located just below or slightly medial to the normal breast tissue.[1] They are histologically distinctive and can contain some or all the components of the nipple and areola.[5] Whilst polythelia usually develop along the mammary lines, they can also occur in atypical locations such as arms/legs, back, shoulders, buttock, neck, vulva, or perineum; however, this is rare.[2] The incidence of polythelia ranges from 0.2% to 5.6% and whilst they are present from birth some are only diagnosed in later life; as is the case of our patient.[1] We present a case of a 42-year-old man who was incidentally found to have a supernumerary nipple on his left buttock.

CASE REPORT

A 42-year-old male was referred from his GP for a colonoscopy, as he was experiencing some per-rectal (PR) bleeding and tenesmus. He has a past medical history of GORD and vitamin D deficiency. He had no other risk factors or family history of colorectal cancer.

Prior to beginning the colonoscopy, the surgeon preformed an external and digital rectal examination (DRE). Whilst the DRE was unremarkable, the surgeon did notice an abnormality to the patients left buttock. In the left gluteal fold, there was a circular hyperpigmented lesion approximately 2 cm in diameter. On closer inspection the lesion was highly suspicions for a nipple, as it was elevated with a central projection and was surrounded with strands of hair (Figure 1). The surrounding tissue was normal and there were no palpable masses or accessory breast tissue (polymastia) associated with the lesion. Grade II internal hemorrhoids were also found at the colonoscopy and thought to be the cause of the patient's PR bleeding. The hemorrhoids were treated with hemorrhoid energy therapy.

At the clinic appointment following his colonoscopy it was discussed with the patient that he had an accessory nipple. The patient was aware of the lesion as it had been there since birth however, he was under the impression that it was a birth mark. On further discussion, the patient did not have any history of urinary tract abnormalities and there was no family history of congenital abnormalities, cancers or polythelia. Additionally, there were no other ectopic nipples or associated breast tissue

found on a thorough examination. The patient decided on conservative management of his polythelia as he is asymptomatic, and it has been present since birth.

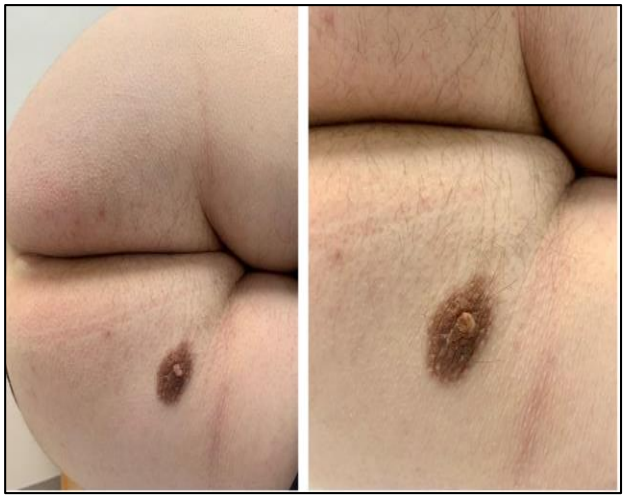

Figure 1: Supernumerary nipple on the patient's left buttock.

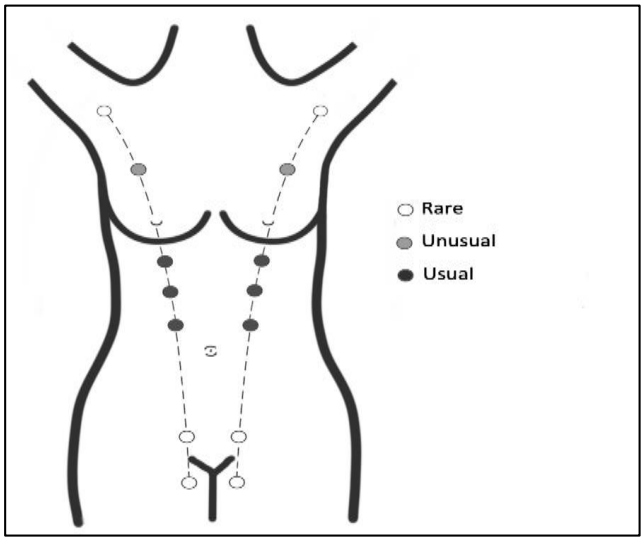

Figure 2: Diagram of the distribution of the mammary "milk" lines and potential sites for accessory nipples and breast tissue.

DISCUSSION

Whilst polythelia is a rare condition it is one of the most common malformations involving mammary tissue.[3] Polythelia or supernumerary nipples are often colloquially referred to as a "third nipple", however the clinical condition is a bit more complex. The classification of polythelia requires a developed areola with a nipple.[4] Interestingly, every supernumerary nipple is associated with an areola however not every supernumerary areola has a nipple.[4]

Most cases of polythelia appear sporadically however there have been some instances of hereditary spread.[3,5] Despite this there has not been any genetic mutations identified for either inheritance method.[6] However, there has been an increased association between polythelia and a number of congenital and chromosomal anomalies in the literature. These anomalies include Char syndrome, Turner's syndrome, arthrogryposis multiplex congenital, fetal alcohol syndrome, neurofibromatosis type 1, Fleischer's syndrome, partial chromosome 3p trisomy, trisomy 8 and Simpson-Galabi-Behmel syndrome.[7-10] While the supernumerary nipple itself is often benign, they have been linked to a higher incidence of isolated urinary tract malformations and urogenital malignancies.[6,11] Other congenital conditions such as malformation to the cardiac, vertebral and central nervous systems, as well as dental anomalies have also been linked to polythelia in the literature.[6]

The link between polythelia and these associated congenital abnormalities is likely hidden in embryonic development. Typically, between the 4th and 5th weeks of gestation the cells of the ectoderm proliferate to form two lines that extend from the fetal axilla to the groin. These primitive lines, known as the milk lines, thicken during the second to third trimester to form the mammary ridges (Figure 2).[5,12,13] As the mammary ridges continue to develop, forming the primary mammary buds in the pectoral region, the remainder of the primitive milk line regresses. However, incomplete or failure of the mammary ridge to regress can result in the development of aberrant mammary tissue, such as supernumerary nipples.[2,5,12]

Table 1: Kajawa's clinical classification of aberrant mammary tissue.

S. no.	Classification
1	Complete breast with nipple, areola, and glandular tissue
2	Supernumerary breast without areola but with nipple and glandular tissue
3	Supernumerary breast without nipple but with areola and glandular tissue
4	Aberrant glandular tissue only without nipple and areola
5	Pseudomamma (nipple and areola with glandular tissue replaced by fat)
6	Polythelia (nipples only)
7	Polythelia areolaris (areola only)
8	Polythelia pilosa (patch of hair only)

Data adapted from Kajava (1915)

Polythelia was first published by Leichtenstern in 1878 however, it is more commonly known via the Kajawa's classification which was later published in 1915 (Table 1).[14] Since then there have been numerous other cases of polythelia published within the literature, however there are no cases of a supernumerary nipple located on the buttocks. This is an unusual location for polythelia to occur as it does not follow the milk lines, but they have

been known to form in other locations.[2] There has been the many theories postulated through the literature for the presence of supernumerary nipples and aberrant mammary tissue outside of the milk lines. One theory is that there is displacement of the mammary crests leading to the formation of supernumerary nipples.[2] Alternatively, Newman (1988) theorized in his publication that supernumerary nipples are just modified apocrine sweat glands and therefore can occur at any location.[15] Some authors have even postulated that polythelia is an atavistic manifestation. One justification is the parallels noted between ectopic mammary tissue sites and the usual location of mammary tissue in other mammals.[2,13]

CONCLUSION

In summary, this is the first published case of a supernumerary nipple on the buttock. Despite multiple case reports that have been published there is limited larger patient studies to better understand polythelia and its clinical significance.

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