CASE REPORTS IN PLASTIC SURGERY AND HAND SURGERY
2025, VOL. 12, NO. 1, 2487843
https://doi.org/10.1080/23320885.2025.2487843

Taylor & Francis
Taylor & Francis Group

RESEARCH ARTICLE

Areolar sebaceous hyperplasia: case report and literature review

Amanda Rekola[a], Daniel Tegnelius[b], Emma Wall[c], Johann Zdolsek[c] and Reza Tabrisi[c,d]

[a]School of Medical Sciences, Örebro University, Örebro, Sweden; [b]Department of General Surgery, Örebro University Hospital, Örebro, Sweden; [c]Department of Head, Neck, and Reconstructive Plastic Surgery, Örebro University Hospital, Örebro, Sweden; [d]Faculty of Medicine and Health, Örebro University, Örebro, Sweden

ABSTRACT

Areolar sebaceous hyperplasia (ASH), also known as Montgomery hyperplasia, is an uncommon benign neoplasm. Despite nearly 40 years since the first case was described, this condition remains rare, with only 20 reported cases. We present a case of ASH in a 44-year-old female and review the existing literature.

ARTICLE HISTORY
Received 30 September 2024
Accepted 29 March 2025

KEYWORDS
Areolar sebaceous hyperplasia; nipple lesion; Montgomery's tubercles; Montgomery hyperplasia; areola; sebaceous neoplasm; NAC

Introduction

Areolar sebaceous hyperplasia (ASH), also known as Montgomery hyperplasia, presents as yellowish plaque-like lesions on the areola and/or nipple, consisting of multiple small papules [1]. Biopsy is recommended, particularly in cases with unclear or related pathology, to rule out the possibility of malignancy [2]. While common diagnostic methods include biopsy and histopathological examination, Cozzolino et al. proposed a noninvasive approach using cytoscraping for diagnosing ASH [3]. Paget's disease of the breast should be considered as a differential diagnosis, particularly in postmenopausal individuals with nipple lesions [2].

Histopathological findings of ASH typically reveal several enlarged mature sebaceous glands located in the upper dermis [2–14]. These glands open either directly or through a short connecting duct to the epidermis [4–18]. In some cases, hair follicles are observed in association with the duct, aligning with the definition of sebaceous hyperplasia [19]. Conversely, studies on Montgomery glands have not detected any associated hair follicles using either light microscopy or ultrasound [20,21].

Since ASH lesions are benign, treatment is mainly performed for cosmetic reasons [18,22,23]. However, surgical excision may be necessary for symptom relief in cases involving sensory discomfort or large tumors. Tulbert and Brodell described a non-surgical removal option using 100% dichloroacetic acid [18]. Since Catalano and Ioannides first reported a case of ASH [5], relatively few cases have been documented in the literature, and no common treatment guidelines are at hand. The aim of our report is to review relevant literature on ASH and, in light of the predominantly non-surgical treatments described, present a case of successful surgical excision. A signed, written informed consent for participation was received from the patient.

Case description

A 44-year-old female patient presented to the outpatient clinic at the Department of Surgery at Örebro University Hospital, Sweden, with a two-year history of a growing lesion on her right nipple. Her symptoms included increasing discomfort, pain, unpleasant odor and discharge from the nipple-areolar complex (NAC). The medical history was unremarkable. On clinical examination, a 4 cm mass was observed on the right NAC (Figure 1(A,B)). The right breast was soft, with no palpable pathological lesions detected. Mammographic and ultrasonic investigations were normal, showing no

CONTACT Reza Tabrisi reza.tabrisi@oru.se Department of Head, Neck, and Reconstructive Plastic Surgery, Örebro University Hospital, Örebro, Sweden
Supplemental data for this article can be accessed online at https://doi.org/10.1080/23320885.2025.2487843.

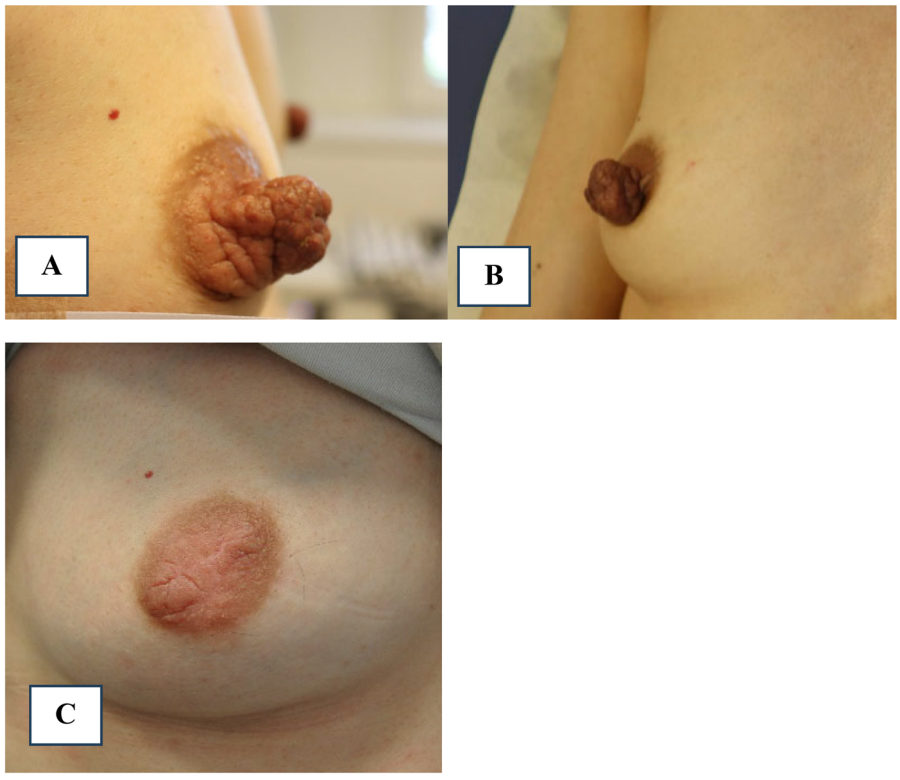

Figure 1. NAC lesion on the right breast at initial presentation (A, B), and one year after surgery (C).

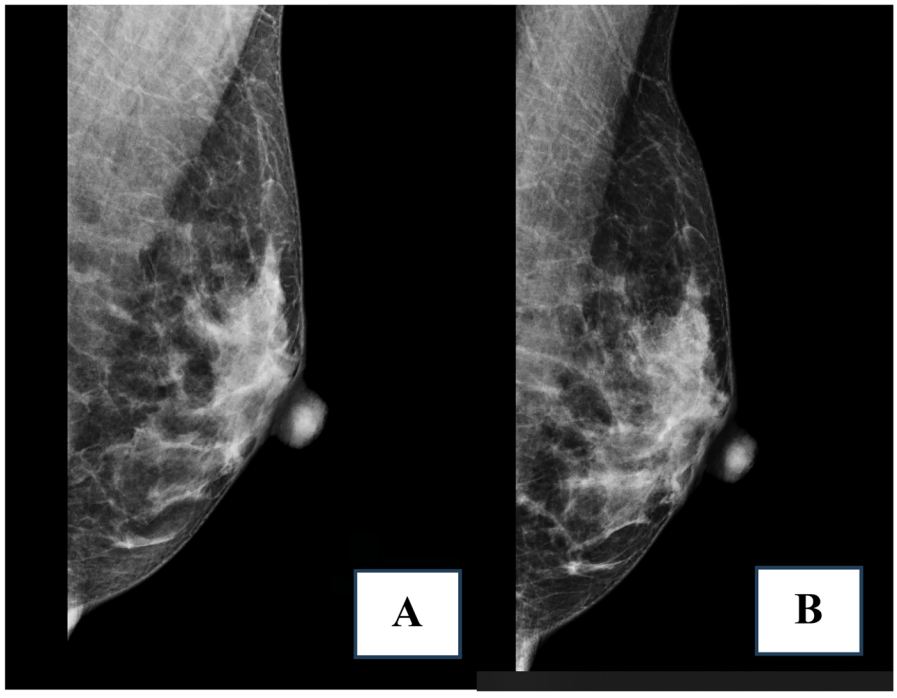

Figure 2. Mammographic illustration at initial presentation (A), and one year after surgery (B).

signs of mammary malignancy (Figure 2(A)). Cytological examination could not be performed as the nipple discharge had ceased. However, a punch biopsy was conducted, which ruled out malignancy.

Histopathological examination revealed a normal to slightly acanthotic epidermis with multiple large ectopic sebaceous glands, or Montgomery's tubercles, directly connected to the epidermis, with some

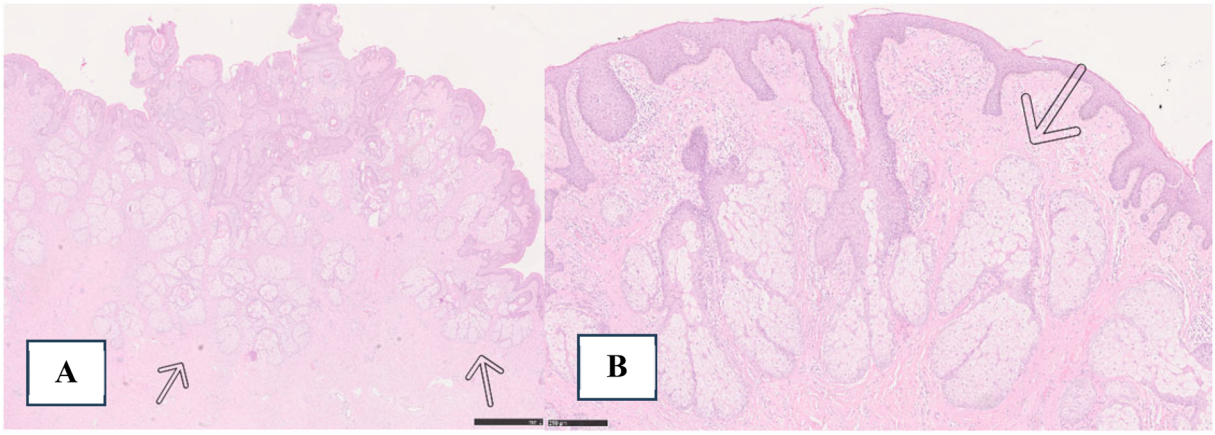

Figure 3. Histopathological evaluation of the patient, arrows indicating Montgomery's tubercles. (A) Magnification, ×2. (B) Magnification, ×10.

opening to the surface (Figure 3). Due to the patient's discomfort and personal preference, surgical excision with primary closure was chosen. As the lesion did not involve the mammary duct (Figure 1(A,B)), no cannulation technique was utilized.

No post-operative complications or adverse events were reported. At the 12-months follow-up, the patient presented with a well-healed minimal scar at the excision site and intact nipple sensitivity (Figure 1(C)). Minor periods of local sensory discomfort were reported, but overall satisfaction was high. A control mammography was conducted at the follow-up showed normal findings (Figure 2(B)).

Literature review

Search strategy

Search terms such as *sebaceous hyperplasia*, *Montgomery's tubercles* or *hyperplasia* are commonly used terms in the literature. Consequently, a search strategy was developed by a certified science librarian. A literature search was conducted in PubMed® and Embase via Elsevier on 17 January 2025, without a specified time frame. Details of the search strategy are provided in Figures 4 and 5.

Eligibility criteria

No language restrictions or time limit were applied. All study types were included in the search. Studies describing human cases were included, while those without human cases were excluded.

Study selection

A total of 229 articles (124 from PubMed and 105 from Embase) were identified through the literature search. After removing duplicates and non-English articles, 131 articles remained. A blinded title and abstract screening for eligibility was conducted by two independent investigators (A.R. and R.T.), resulting in the exclusion of 86 articles. A second assessment involving a blinded full text screening was performed by the investigators excluding 25 records in total. Following a full-text screening excluded additional 25 articles in total. Following a full-text screening, ultimately 20 articles were deemed relevant and included in this study, as outlined in Table 1.

Discussion

ASH is an uncommon yet benign tumor of the NAC and can present unilaterally (n = 10) or bilaterally (n = 11), with female cases reported twice the rate of males [3–19,25–27]. With only 21 reported cases, it is premature to establish definitive patterns regarding age, gender and anatomical distribution of Montgomery's tubercles on the NAC (Table 2).

Current literature offers conflicting views on the morphological characteristics of Montgomery's tubercles in males and females, as well as their association with ASH. Krause suggests that these structures exist in both genders, with sebaceous glands in males and accessory lactiferous glands and ducts in females [22]. Conversely, some studies argue that men do not have Montgomery's tubercles [7,11]. While some studies indicate no association between ASH and Montgomery's tubercles [4,14], others have reported cases where these tubercles were present [23,25]. In this specific case, hyperplasia of Montgomery's tubercles was observed during histopathological examination.

The etiology of ASH remains unclear, although hormonal influence may play a role in its pathophysiology. Two previously documented cases linked ASH

	PubMed® Search terms	Number of results
Montgomery´s tubercles		
1	(Montgomery*[Title/Abstract] AND (gland*[Title/Abstract] OR tubercle*[Title/Abstract])) OR ("Areolar gland*"[Title/Abstract] OR "Aerolar gland*"[Title/Abstract] OR "glandulae areola*"[Title/Abstract] OR "Tubercula areola*"[Title/Abstract])	46
Nipple (location where the problem might occur)		
2	"Nipples"[Mesh] OR nipple*[Title/Abstract] OR areola*[Title/Abstract] OR aerola*[Title/Abstract]	15,316
Sebaceous hyperplasia		
3	sebaceous[Title/Abstract] OR "Sebaceous Glands"[Mesh]	12,11
4	Hyperplasia*[Title/Abstract] OR "Hyperplasia"[Mesh]	116,105
5	#3 AND #4	761
Choristoma		
6	"Choristoma"[Mesh] OR Choristoma*[Title/Abstract] OR "Ectopic Tissue*"[Title/Abstract] OR "Heterotopic Tissue*"[Title/Abstract] OR "Aberrant Tissue*"[Title/Abstract]	9,841
Sebaceous hyperplasia OR Choristoma		
7	#5 OR #6	10,591
Sebaceous hyperplasia OR Choristoma combined with location (nipple)		
8	#7 AND #2	80
All sets combined		
9	#1 OR #8	124

	Embase Search terms	Number of results
Montgomery´s tubercles		
1	montgomery*:ti,ab,kw AND (gland*:ti,ab,kw OR tubercle*:ti,ab,kw) OR 'areolar gland*':ti,ab,kw OR 'aerolar gland*':ti,ab,kw OR 'glandulae areola*':ti,ab,kw OR 'tubercula areola*':ti,ab,kw	63
Nipple (location where the problem might occur)		
2	'nipple'/de OR 'breast areola'/de	9897
3	nipple*:ti,ab,kw OR areola*:ti,ab,kw OR aerola*:ti,ab,kw	18341
4	#2 OR #3	20377
Sebaceous hyperplasia		
5	'sebaceous gland hyperplasia'/de	311
6	'sebaceous gland'/de	5858
7	sebaceous:ti,ab,kw	11458
8	#6 OR #7	13506
9	hyperplasia*:ti,ab,kw	143192
10	#8 AND #9	960
11	#5 OR #10	1084
Choristoma		
12	'choristoma'/de	2969
13	choristoma*:ti,ab,kw OR 'ectopic tissue*':ti,ab,kw OR 'heterotopic tissue*':ti,ab,kw OR 'aberrant tissue*':ti,ab,kw	2623
14	#12 OR #13	4568
Sebaceous hyperplasia OR Choristoma		
15	#11 OR #14	5648
Sebaceous hyperplasia OR Choristoma combined with location (nipple)		
16	#4 AND #15	44
All sets combined		
17	#1 OR #16	105

Figure 4. Search terms and number of results.

development to pregnancy and lactation [5,8]. Additionally, sebaceous hyperplasia in newborns has been attributed to maternal hormonal influence [26]. Sebocytes express androgen receptors and may proliferation in response to elevated androgen levels [27]. A recent study by Oya et al. highlighted the

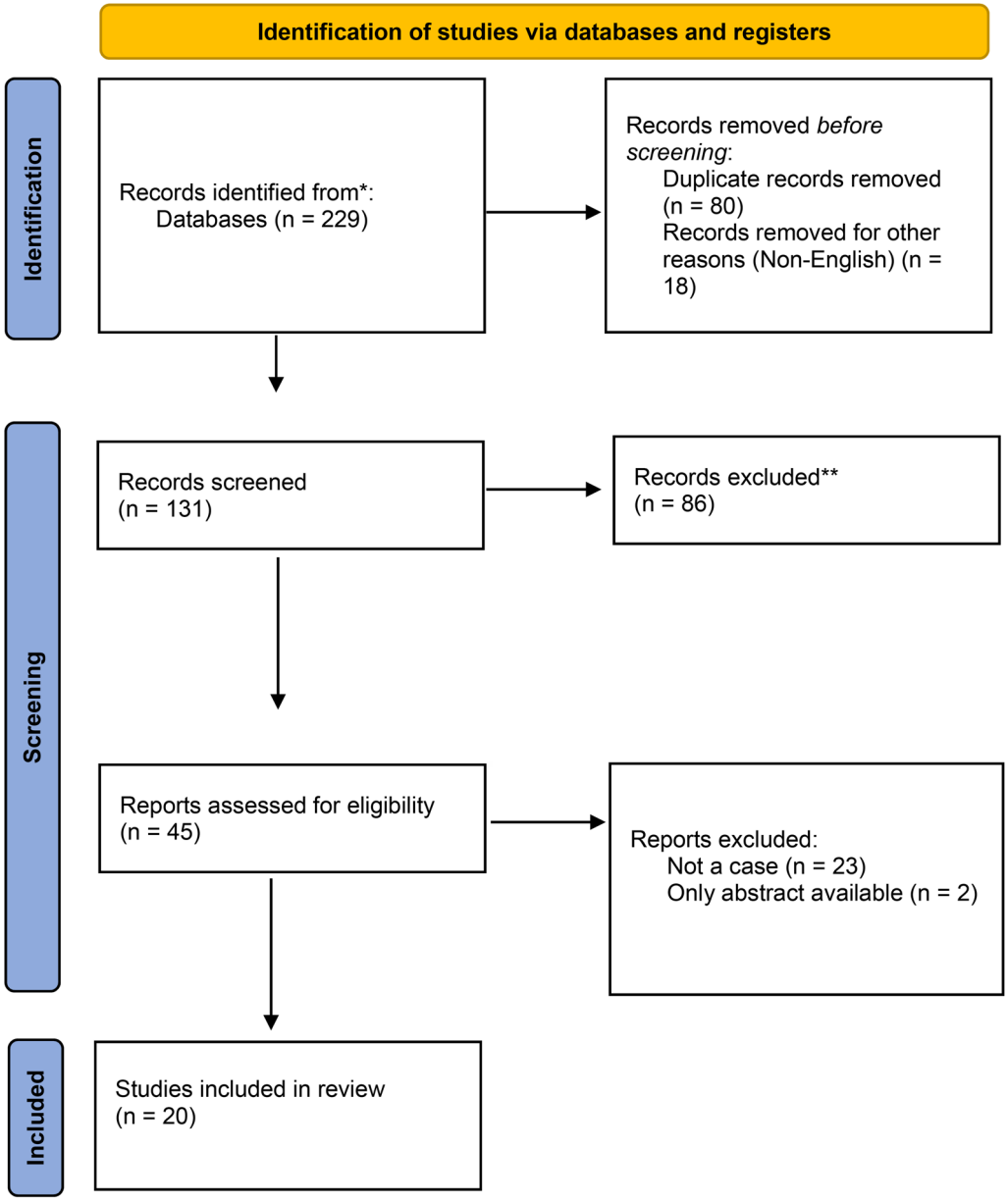

Figure 5. Identified cases with Montgomery's tubercles out of 229 identified records. *Identified 124 records on PubMed® and 105 records on Embase. **No automation tools were used to identify cases; all cases were identified by the authors.

expression of androgen receptors in sebocytes, further supporting their role in the development of ASH [12].

Excisional surgery has been recommended to provide symptom relief. In this particular case, the patient expressed sensory discomfort and concerns about the increased size of the lesion among other symptoms. Additionally, she desired a full NAC-excision to ensure complete removal of the lesion. Studying the literature, no common guidelines were found with regards to treatment strategies for ASH. As demonstrated in Table 1, treatment strategies varied widely. Seven cases employed conservative management, two used topical treatments, one involved mastectomy due to ipsilateral carcinoma, and another described total NAC excision. In nine of the articles, there was no report on chosen treatment (Table 1). After a departmental review of the literature and the patient's case, surgical excision of the lesion alone was recommended and performed. At the 12-month follow-up, there were no signs of recurrence, and the patient expressed satisfaction with the cosmetic results and retained sensation. Given the disturbing nature and localization of ASH lesions, we propose that surgical removal should be generously offered to affected patients.

Table 1. Literature review: cases chronologically by year.

Case	Study	Sex	Location	Morphology	Age	Treatment strategy	Comments
1	Catalano and Ioannides [5]	F	Bilateral (A)	Plaque, small dots	37	Missing	After childbirth
2	Sánchez Yus et al. [13]	F	Unilateral (A)	Single papule	59	Excision	Diagnosed as xanthoma before excision.
3	Hammerton and Shrank [9]	F	Bilateral (A + N)	Several papules		Missing	Sore nipples. Fordyce spots on upper lip. Benign breast nodule. Hysterectomized.
4	Tsuji and Yamauchi [14]	F	Bilateral (A)	Plaque	43	Local steroids	Leukorrhea, irregular menstrual cycle.
5	Fariña et al. [7]	M	Unilateral (A)	Thickening, papillary surface	49	Missing	Hypogonadism
6	Belinchón et al. [4]	F	Bilateral (A)	Plaque	42	Missing	
7	Kondo-Morita et al. [10]	M	Bilateral (A)	Thickening	37	Missing	Yellowish spots on buccal mucosa.
8	Krisp and Krause [11]	M	Bilateral (A)	Thickening, papillary surface	52	Missing	
9	Bajaj et al. [24]	M	Unilateral (A)	Plaque	49	No treatment	
10	Conde-Taboada et al. [6]	M	Unilateral (A)	Plaque, papillated surface.	24	Missing	
11	Guillermo et al. [17]	F	Bilateral (A)	Plaque	43	No treatment	Pruritus. Fordyce spots on upper lip. Previous breast fibroadenoma. Dyslipidemia
12	Gao et al. [8]	F	Unilateral (A + N)	Papules, nodules. Papillated surface.	37	No treatment	After pregnancy and lactation
13	Tulbert and Brodell [18]	F	Unilateral (A)	Papules	15	Local dichloroacetic acid	Thermal injury as a child
14	Fernandez-Flores et al. [16]	F	Unilateral (A)	Thickened plaque	32	Mastectomy	Donohue syndrome, ipsilateral invasive ductal carcinoma
15	Errichetti et al. [15]	M	Bilateral (A)	Plaque, papillomatous surface		No treatment	Fordyce spots on upper lip and penis shaft
16	Lester et al. [23]	F	Bilateral (A + N)	Thickened plaque	23	No treatment	Oily discharge
17	Cozzolino et al. [3]	F	Unilateral (N)	Plaque	50	Missing	
18	Chiriac et al. [2]	F	Bilateral (A)	Plaque with smooth/ papillary surface	67	No treatment	Post-menopausal
19	Oya et al. [12]	M	Unilateral (A)	Nodular	41	No treatment	Areolar leiomyoma
20	Kelly et al. [25]	F	Bilateral (A + N)	Coalescing plaque	30	Missing	

F: female; M: male. A: areola; N: nipple.
Age, gender, location and morphology of lesion with additional comments for each case are described.

Table 2. Patient characteristics: number of cases (%), Age ± SD, (range) and lesion location (%).

	Female	Male
Number of cases	14 (67%)	7 (33%)
Age	39.8 ± 14.5	42.0 ± 10.5
Range	(15–67)	(24–52)
Location		
Unilateral	6 (43%)	4 (57%)
Bilateral	8 (57%)	3 (43%)
Areola	8 (57%)	7 (100%)
Nipple	1 (7%)	–
Areola + nipple	5 (36%)	–

Conclusions

In summary, the etiology of ASH, also known as Montgomery's hyperplasia, remains unidentified, although the condition appears to be benign. Montgomery tubercles are located beneath the areola. Although they do not contain ducts, hyperplasia may affect the ducts. Most published cases have been treated conservatively, with only a few managed through surgical excision. No established consensus exists on the best treatment regimen. Additionally, surgical treatment may impact the ducts and potentially disrupt lactation. In patients with expectations of having children and a desire to breastfeed, could conservative management be preferred since ASH is a benign lesion.

Here, we report on a case of a 44-year-old female with a unilateral 4 cm ASH lesion successfully treated with surgery, resulting in retained nipple sensation. Although long-term follow-up data on conservatively treated ASH are lacking, we recommend surgical excision, as the lesions are unlikely to regress spontaneously. Surgical excision offers a definitive solution.

Acknowledgements

We extend our deepest gratitude to Dr. Ghaboosi for her expert language revision.

Disclosure statement

No potential conflict of interest was reported by the author(s).

Funding

We hereby declare that there is no financial gain associated with any organization regarding the contents of this manuscript.

Data availability statement

We hereby confirm that no new data were created or analyzed in this study and that data sharing is not applicable for the contents of this paper.

References

[1] Lin E-Y, Rao L, Wang W-J, et al. Detection of sebaceous gland hyperplasia with dermoscopy and reflectance confocal microscopy. Front Med. 2023;10:1194748. doi: 10.3389/fmed.2023.1194748.

[2] Chiriac A, Moldovan C, Coros MF, et al. Bilateral areolar sebaceous hyperplasia in a post-menopausal woman. Eur J Dermatol. 2016;26(3):299–300. doi: 10.1684/ejd.2016.2739.

[3] Cozzolino I, Thomas G, Palombini L. Unilateral areolar sebaceous hyperplasia on nipple scraping sample. Diagn Cytopathol. 2015;43(5):388–389. doi: 10.1002/dc.23181.

[4] Belinchón I, Aguilar A, Tardío J, et al. Areolar sebaceous hyperplasia: a case report. Cutis. 1996;58(1):63–64.

[5] Catalano PM, Ioannides G. Areolar sebaceous hyperplasia. J Am Acad Dermatol. 1985;13(5 Pt 2):867–868. doi: 10.1016/s0190-9622(85)80270-x.

[6] Conde-Taboada A, De la Torre C, Mayo E, et al. Unilateral areolar sebaceous hyperplasia in a male. J Eur Acad Dermatol Venereol. 2007;21(1):120–121. doi: 10.1111/j.1468-3083.2006.01806.x.

[7] Fariña MC, Soriano ML, Escalonilla P, et al. Unilateral areolar sebaceous hyperplasia in a male. Am J Dermatopathol. 1996;18(4):417–419. doi: 10.1097/00000372-199608000-00016.

[8] Gao J, Liu L, Li C, et al. Premature sebaceous hyperplasia of nipple and areola. Int J Dermatol. 2010;49(7):801–802. doi: 10.1111/j.1365-4632.2009.04440.x.

[9] Hammerton MD, Shrank AB. Superficial sebaceous hyperplasia of the areolae. Br J Dermatol. 1993;129(5):649–650. doi: 10.1111/j.1365-2133.1993.tb00511.x.

[10] Kondo-Morita A, Murata S, Murakami T, et al. Bilateral areolar sebaceous hyperplasia in a male. J Dermatol. 2001;28(3):172–173.

[11] Krisp A, Krause W. Areolar sebaceous hyperplasia. Acta Derm Venereol. 2003;83(1):61–62. doi: 10.1080/00015550310002792.

[12] Oya K, Nakamura Y, Fujisawa Y. Unilateral areolar leiomyoma with sebaceous hyperplasia. Indian J Dermatol Venereol Leprol. 2022;88(4):536–540. doi: 10.25259/IJDVL_7_2020.

[13] Sánchez Yus E, Montull C, Valcayo A, et al. Areolar sebaceous hyperplasia: a new entity? J Cutan Pathol. 1988;15(1):62–63. doi: 10.1111/j.1600-0560.1988.tb00519.x.

[14] Tsuji T, Yamauchi R. Areolar sebaceous hyperplasia with a Fordyce's spot-like lesion. J Dermatol. 1994;21(7):524–526. doi: 10.1111/j.1346-8138.1994.tb01787.x.

[15] Errichetti E, Piccirillo A, Viola L, et al. Areolar sebaceous hyperplasia associated with oral and genital Fordyce spots. J Dermatol. 2013;40(8):670. doi: 10.1111/1346-8138.12191.

[16] Fernandez-Flores A, Valerdiz S, Crespo LG, et al. Areolar sebaceous hyperplasia with underlying primary duct carcinoma of the breast in a woman with Donohue syndrome (leprechaunism). Am J Dermatopathol. 2012;34(2):e15–e18. doi: 10.1097/DAD.0b013e318231311a.

[17] Guillermo N, Peñate Y, Soler E, et al. Bilateral areolar sebaceous hyperplasia in a female. Int J Dermatol. 2008;47(11):1214–1215. doi: 10.1111/j.1365-4632.2008.03856.x.

[18] Tulbert B, Brodell RT. A simple and effective treatment for ectopic sebaceous glands on the areola. Dermatol Surg. 2010;36(8):1332–1335. doi: 10.1111/j.1524-4725.2010.01634.x.

[19] Papadimitriou I, Vakirlis E, Sotiriou E, et al. Sebaceous neoplasms. Diagnostics. 2023;13(10):1676. doi: 10.3390/diagnostics13101676.

[20] Wortsman X, Carreño L, Ferreira-Wortsman C, et al. Ultrasound characteristics of the hair follicles and tracts, sebaceous glands, Montgomery glands, apocrine glands, and arrector pili muscles. J Ultrasound Med. 2019;38(8):1995–2004. doi: 10.1002/jum.14888.

[21] Smith DMJr., Peters TG, Donegan WL. Montgomery's areolar tubercle. A light microscopic study. Arch Pathol Lab Med. 1982;106(2):60–63.

[22] Krause W. Diseases of the male nipple and areola. J Dtsch Dermatol Ges. 2011;9(12):1004–1009. doi: 10.1111/j.1610-0387.2011.07720.x.

[23] Lester RA, Torgerson RR, Sandhu NP. Rare presentation of sebaceous hyperplasia. BMJ Case Rep. 2014;2014:bcr2014204025. doi: 10.1136/bcr-2014-204025.

[24] Bajaj V, Barrett P, Sripathy T, et al. Areolar sebaceous hyperplasia in a male -- a different morphology. J Cutan Pathol. 2007;34(2):207–208.

[25] Kelly K, Weir K, Gardner R. Areolar sebaceous gland hyperplasia. BMJ Case Rep. 2023;16(12). doi: 10.1136/bcr-2023-258492.

[26] Krüger EMM, Sinkos F, Uhry JF, et al. Dermatoses in the early neonatal period: their association with neonatal, obstetric and demographic variables. Rev Paul Pediatr. 2019;37(3):297–304. doi: 10.1590/1984-0462/;2019;37;3;00012.

[27] mZouboulis CC. Sebaceous gland receptors. Dermatoendocrinol. 2009;1(2):77–80. doi: 10.4161/derm.1.2.7804.