

Management of Congenital Melanocytic Naevi in Children: A French National Survey Using Clinical Vignettes

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Management of congenital melanocytic naevi in childhood may vary depending on the habits and experience of the treating clinician. The aim of this study was to assess current practice and determinants of surgical excision decision-making among French physicians. A national survey was conducted among dermatologists, paediatricians and surgeons, using clinical vignettes illustrating 29 scenarios. The primary outcome was the decision to perform surgical excision in each vignette. Of the 11,310 decisions made by the 390 participants (257 dermatologists, 35 surgeons, and 98 paediatricians) surgical excision was chosen in 33% of cases. The stated motivations for performing surgical excision were: melanoma risk, aesthetic/psychosocial risk, or both, in 39%, 34% and 27% of cases, respectively. Physicians with a higher level of experience in oncodermatology were more likely to opt for surgical excision. The age of the child, the size of the congenital melanocytic naevi, and the visibility of the lesion had no influence on the decision to perform surgical excision.

Key words: congenital naevus; melanoma; congenital melanocytic naevi; naevi/therapeutic.

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Congenital melanocytic naevi (CMN) are a benign proliferation of melanocytes present at birth or in the first weeks of life, presenting with single or multiple cutaneous lesions, ranging from light-brown to dark-brown, sometimes bluish, with or without hypertrichosis. Recently, multiple CMNs have been associated with post-zygotic mutations in the *NRAS* gene (1) Depending on the timing of these mutations during embryogenesis, CMNs may sometimes be associated with extracutaneous symptoms as part of what is described as “CMN syndrome” (2) including neurocutaneous melanosis (NCM) (3).

CMN classification was reviewed by Krengel in 2012 (4). This classification is based mainly on the projected adult size (PAS) of the CMN. The CMN PAS categories are: small (<1.5 cm); medium (1.5–20 cm); large (>20–40 cm); and giant (>40 cm). In addition, the number of satellite naevi in the first year of life is categorized into

SIGNIFICANCE

Despite the frequency of congenital melanocytic naevi, their therapeutic management does not usually involve shared decision-making, and may vary depending on the habits and experience of each clinician. This survey reveals the variability of management of congenital melanocytic naevi among clinicians at a national level in France. Management varied depending on: the clinician’s level of experience in oncodermatology; their previous management of a melanoma on congenital melanocytic naevi; their seniority; and the case-load mix. National recommendations to standardize clinical practice and the information provided to patients should be developed to help guide a shared decision-making process.

none, 1–20, more than 20–50, and more than 50 satellites. Additional descriptors of CMN include anatomical localization, colour heterogeneity, surface rugosity, presence of hypertrichosis, and presence of dermal or subcutaneous nodules.

CMN are common lesions, although their precise prevalence is not known, it estimated as between 1% and 3% of newborns (3–6). Most lesions are small. The incidence of large or giant CMN is lower; between 1:20,000 and 1:500,000 births (7, 8).

The 3 main issues associated with CMN are the risk of malignant transformation into melanoma throughout life, the psycho-social and cosmetic burden, and the risk of CMN neurological complications.

The decision to perform surgical excision of CMN is often based on melanoma risk. Although complete surgical excision eliminates the lifelong risk of melanoma for small, and sometimes for medium, lesions, this is not the case for large/giant CMN. Although surgical excision may reduce the psychosocial and cosmetic impact of CMN, the benefit/harm ratio should be assessed on an individual basis, depending on the localization, age and size of the lesion.

Despite their frequency, therapeutic management of CMN does not usually involve shared decision-making and may differ depending on the habits and experience of each clinician.

The aim of this study was to assess: (i) current national practices regarding the therapeutic management of CMN (surgical excision vs abstention) by dermatologists, pa-

ediatricians and surgeons; and (ii) the determinants of decision-making according to the physicians' characteristics (age, specialty, adult vs paediatric practice, level of experience in oncodermatology) and to the CMN characteristics (size, location, age of the child). The study hypotheses were that: (i) a higher level of experience in oncodermatology, including management of melanomas on CMN in adults, would increase the likelihood of opting for surgical excision in children with CMN; (ii) the size and location of the CMN would be determinants for decision-making regarding surgical removal.

METHODS

A pilot-tested questionnaire containing a range of clinical vignettes was developed online. The vignettes illustrated clinical scenarios, in which the age of the child and classification of the CMN (localization, size) varied. Between December 2018 and March 2019 the questionnaire was sent by e-mail to all members of academic groups of French dermatologists, paediatricians, surgeons (plastic and paediatric [Academic groups: Société française de dermatologie; Association française des pédiatres ambulatoires; Société française de pédiatrie; Société française de chirurgie plastique pédiatrique.]). The vignettes were presented in a random order to each participant.

The primary outcome was the decision to perform surgical excision (yes/no) for each vignette. In case of surgical decision, the physicians were asked about their motivation (cosmetic/psychosocial risk, melanoma risk, or both). In case of surgical abstention, the physicians were asked whether clinical follow-up was needed and at what age this should take place. Potential determinants for surgical excision studied were the physicians' characteristics (including their experience of managing malignant transformation of a CMN) and the clinical characteristics of the cases: patient's age (under 3 years, 3–10 years, and over 10 years), location on a visible or non-visible area when dressed and PAS (small, medium, large or giant). A total of 29 vignettes illustrating each combination of clinical determinants (varying patient age and lesion location and size) were created. For medium size CMN, an additional determinant was added (technical ease for surgical excision).

This study was approved by the ethics committee of Nantes University Hospital. Parental consent was obtained to use photographs of their child for this study. Univariate and multivariate mixed effects logistic models with random effect on physician identifier were used to identify determinants for decision to perform surgical excision, and χ^2 tests were used to analyse the effect of physician specialty or oncodermatology activity on follow-up. Fleiss kappa test was used to analyse the level of agreement between participants.

RESULTS

Participants

A total of 390 physicians (280 female, 71.8%), including 257 dermatologists, 33 plastic surgeons, 2 paediatric sur-

geons, and 98 paediatricians, answered the questionnaire. Mean \pm standard deviation (SD) age was 50 ± 11.34 years.

Among them, 155 clinicians (39.7%) had a mixed case-load (adult and paediatric) with 99 (25.4%) exclusively involved in paediatric practice. In total, 141 clinicians (36.1%) had previously managed malignant transformation of a CMN (Table I).

Concerning the dermatologists, 196 (76.3%) were women, 24.9% worked at hospital, 55.2% in private practice, and 19.8% working in both care settings.

Rate of decision to perform surgical excision

Among a total of 11,310 decisions from 390 participants, surgical excision was opted for in 33% of cases. Of the 29 vignettes, the number of surgical excision decisions per participant ranged from 1 to 29. The motivations declared surgical excision were aesthetic/psychosocial risk, melanoma risk, or both, in 34%, 39%, and 27% of cases, respectively. When the lesion was not removed, follow-up was opted for in 83% of cases, to occur annually from childhood in 71% of cases.

The clinical scenarios associated with more decisions to perform surgical excision were:

- for all physicians: medium CMN of the thigh in a 2-month-old girl (Fig. 1)
- for plastic surgeons: there was the same level of excision in these following 3 cases ($n=26$): medium CMN of the neck in a 11-year-old girl, medium CMN of the scalp, medium congenital naevus of the eyebrow in a 7-year-old boy

Table I. Participants' characteristics

	Surgeon <i>n</i> = 35	Dermatologist <i>n</i> = 257	Paediatrician <i>n</i> = 98	Total <i>n</i> = 390
Sex, <i>n</i> (%)				
Male	24 (68.57)	61 (23.74)	25 (25.51)	110 (28.21)
Female	11 (31.43)	196 (76.26)	73 (74.49)	280 (71.79)
Age, years, <i>n</i>	33	224	80	337
Missing, <i>n</i>	2	33	18	53
[Min-max]	[33.00;70.00]	[26.00;75.00]	[29.00;70.00]	[26.00;75.00]
Mean \pm standard deviation	48.00 \pm 9.05	49.38 \pm 11.66	52.69 \pm 10.94	50.03 \pm 11.34
Clinical practice, <i>n</i> (%)				
Adult and children	11 (31.43)	143 (55.64)	1 (1.02)	155 (39.74)
Adults only	0 (0.00)	3 (1.17)	0 (0.00)	3 (0.77)
Children only,	3 (8.57)	1 (0.39)	95 (96.94)	99 (25.38)
Mainly adults	17 (48.57)	96 (37.35)	0 (0.00)	113 (28.97)
Mainly children	4 (11.43)	14 (5.45)	2 (2.04)	20 (5.13)
Practice setting, <i>n</i> (%)				
At hospital	13 (37.14)	64 (24.90)	14 (14.29)	91 (23.33%)
Private,	15 (42.86)	142 (55.25)	71 (72.45)	228 (58.46)
Both	7 (20.00)	51 (19.84)	13 (13.27)	71 (18.21)
Oncodermatology activity, <i>n</i> (%)				
Quite important	25 (71.43)	166 (64.59)	2 (2.04)	193 (49.49)
Non-existent to low	8 (22.86)	63 (24.51)	96 (97.96)	167 (42.82)
Majority	2 (5.71)	28 (10.89)	0 (0.00)	30 (7.69)
Seniority, <i>n</i>				
[Min-max]	[5.00;40.00]	[2.00;42.00]	[1.00;41.00]	[1.00;42.00]
Mean \pm standard deviation	18.71 \pm 9.04	20.22 \pm 11.00	22.35 \pm 10.60	20.62 \pm 10.77
Already diagnosed or managed a melanoma on a congenital melanocytic naevi, <i>n</i> (%)				
Yes	24 (68.57)	115 (44.75)	2 (2.04)	141 (36.15)
No	11 (31.43)	142 (55.25)	96 (97.96)	249 (63.85)



Fig. 1. Medium congenital naevus on the thigh in a 2-month-old girl.



Fig. 2. Medium congenital naevus on the sole in a 6-year-old boy.

- for dermatologists and for paediatricians: medium CMN of the thigh in a 2-month-old girl.

The clinical scenarios associated with the least surgical excision decisions were:

- for all physicians: a small CMN of the cheek in girl over 10 years old
- for plastic surgeons: a small CMN of the leg in a 9-month-old child
- for dermatologists: medium CMN of the sole in a 6-year-old boy (Fig. 2)
- for paediatricians: giant CMN in a 3-year-old boy.

Determinants for surgical excision decision

In univariate analysis paediatricians performed fewer excisions than dermatologists (odds ratio (OR) 0.59, $p < 0.0001$), with physicians working in exclusively paediatric practice performing fewer excisions than physicians with a mixed caseload (OR 0.59, $p < 0.0001$). (Table II). For one additional year of seniority, the probability of opting for excision increases significantly (OR 1.01, $p = 0.0308$).

Physicians with no/minimal experience in oncodermatology performed less excision than physicians with an extensive experience (OR 0.66, $p = 0.021$). Physicians with experience of managing a case of melanoma on CMN performed more

excisions than those who did not (OR 1.56, $p < 0.0001$). On the other hand, the age of the child, the size of the CMN, and the visibility of the lesion had no influence on the decision to perform surgical excision.

In multivariate analysis, only the specialty and seniority of the physician's practice had a significant effect on decision-making regarding excision (Table III).

Concerning giant CMN under 3 years of age, surgical excision was opted for in 32.6% of cases, for psychosocial risk or melanoma risk in, respectively, 31% and 25.6% of cases. The excision was recommended to be total in 81.4% of cases. Follow-up was advised by 83.1% of participants, with 71.4% of these opting for follow-ups to occur annually during childhood.

Table II. Determinants for decision to perform surgical excision: univariate analysis

Variable	n	OR (95% CI)	p-value
1. Sex, female vs male	11,310	0.67 (0.55; 0.82)	0.0001***
2. Age	9,773	1.01 (1.00; 1.02)	0.1416
3. Speciality	11,310		
Surgeon vs dermatologist		3.87 (2.93; 5.12)	<0.0001***
Paediatrician vs dermatologist		0.59 (0.46; 0.74)	<0.0001***
4. Type of clinical practice:	11,310		
Only adults vs adults and children		0.62 (0.41; 0.95)	0.0263*
Only children vs adults and children		0.59 (0.45; 0.78)	0.0001***
Mainly adults vs adults and children		1.09 (0.87; 1.37)	0.4672
Mainly children vs adults and children		1.18 (0.81; 1.71)	0.3884
5. Place	11,310		
Hospital vs both		1.27 (0.93; 1.75)	0.1352
Private vs both		1.10 (0.83; 1.45)	0.5062
6. Oncodermatology activity	11,310		
Quite important vs majority		0.95 (0.68; 1.34)	0.7720
Non-existent to low vs majority		0.66 (0.46; 0.94)	0.0210*
7. Seniority	11,310	1.01 (1.00; 1.02)	0.0308*
8. History of diagnosed or managed a melanoma on a CMN: yes	11,310	1.56 (1.28; 1.91)	<0.0001***
9. Age of patients	10,530		
<3 vs >10 years		1.01 (0.93; 1.11)	0.7527
3-10 vs >10 years		1.03 (0.94; 1.13)	0.5332
10. Location: visible vs non-visible	10,530	1.00 (0.92; 1.08)	0.9325
11. Size	10,530		
Giant vs small		1.04 (0.90; 1.19)	0.6269
Large vs small		0.99 (0.89; 1.10)	0.8314
Medium (complex removal) vs small		1.07 (0.95; 1.20)	0.2548
Medium (easy removal) vs small		1.08 (0.97; 1.21)	0.1569

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.
OR: odds ratio; 95% CI: 95% confidence interval; CMN: congenital melanocytic naevi.

Table III. Determinants for decision to perform surgical excision: multivariate analysis

	n	OR (95% CI)	p-value
3. Speciality	11,310		
Surgeon vs dermatologist		3.99 (3.01; 5.29)	<0.0001*
Paediatrician vs dermatologist		0.57 (0.45; 0.71)	<0.0001*
7. Seniority		1.02 (1.01; 1.02)	0.0007*

*p < 0.001.
OR: odds ratio; 95% CI: 95% confidence interval.

Table IV. Proportion of follow-up decision in case of surgical abstention

	Surgeon	Dermatologist	Paediatrician	Total	p-value
Follow-up, n	389	5,355	2,311	8,055	<0.0001
Yes, n (%)	355 (91.26)	4,272 (79.78)	2,079 (89.96)	6,706 (83.25)	
No, n (%)	34 (8.74)	1,083 (20.22)	232 (10.04)	1,349 (16.75)	

Table V. Influence of oncodermatology activity on follow-up decision

	No n = 1,349	Yes n = 6,706	Total n = 8,055	p-value
Oncodermatology activity among dermatologist and surgeons, n	1,117	4,627	5,744	<0.0001
Missing, n	232	2,079	2,311	
Quite important, n (%)	731 (65.44)	3,063 (66.20)	3,794 (66.05)	
Non-existent to low, n (%)	214 (19.16)	1,165 (25.18)	1,379 (24.01)	
Majority, n (%)	172 (15.40)	399 (8.62)	571 (9.94)	

In case of surgical abstention, specialty had a significant effect on the follow-up decision (Table IV). Follow-up was more frequently opted for by surgeons and paediatricians (91% and 90%) than by dermatologists (80%) (p < 0.0001).

Moreover, the level of experience in oncodermatology had a significant effect on the follow-up decision (Table V). Physicians with no/minimal experience in oncodermatology advised, on average, more follow-ups than those with a higher level of experience.

Heterogeneity of decision between specialties and among dermatologists

There was a lack of concordance between participants ($\kappa = -0.893$ (p = 0.895)), and among dermatologists $\kappa = -0.000468$ (p = 0.648) (Fig. 3).

DISCUSSION

This survey on the management of CMN in France, using clinical vignettes, highlights a high degree of heterogeneity of declared practices, both between physician specialties (dermatology, paediatrics, surgery) as

well as within them (dermatology). Decisions for CMN excision were as much attributed to the risk for melanoma as to the aesthetic and psychosocial risk. As expected, univariate analysis showed physicians with an extensive level of experience in oncodermatology were more likely to opt for surgical excision than those without such experience. Younger physicians, paediatricians and physicians who worked exclusively with paediatric patients were less likely to opt for surgical excision. Thus, melanoma risk is probably perceived more by dermatologists than paediatricians, and among dermatologists who have a higher level of experience in oncodermatology. Contrary to what we had expected, neither patient age, size, or location of the CMN were found to be determinant factors for surgical excision in this study.

The precise magnitude of melanoma risk associated with CMN is not known. From published data, this risk is estimated as between 0.7% and 2%, and up to 10% for giant CMN (9–11). This risk is probably low for small- and medium-size CMN, but data to inform such estimates are too scarce to be definitive. In a follow-up study of 230 lesions in 227 patients from Sahin et al. (12), no melanomas arose in any medium-sized CMN during a mean follow-up of 6.7 years, up to a mean age of 25.5 years. We do, however, know that this risk is increased by the size of the CMN, the number of CMN and satellites, as well as the involvement of the central nervous system.

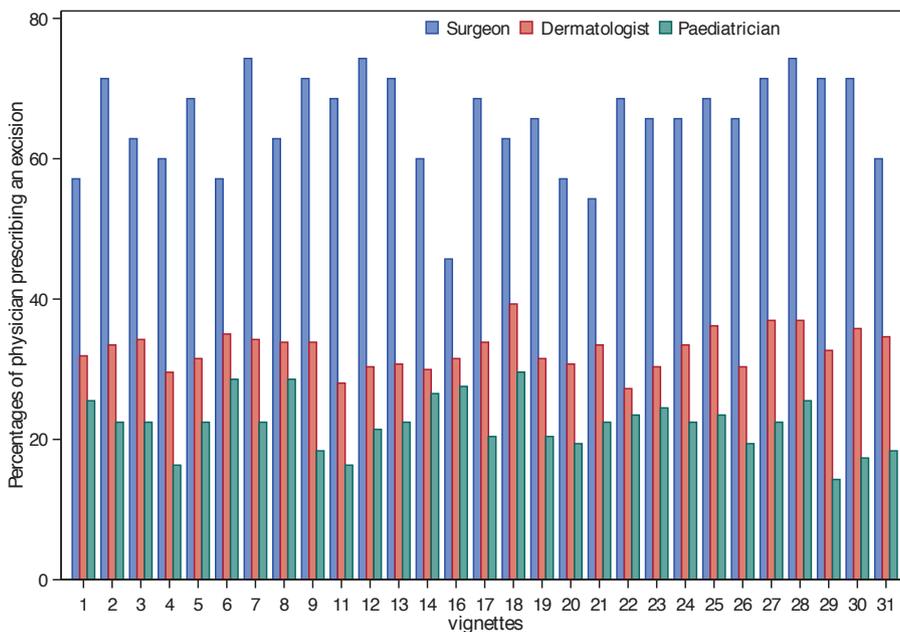


Fig. 3. Heterogeneity of prescribing of events among disciplines and dermatologists according to vignettes.

Finally, on the molecular level, it is now known that the presence of multiple CMN (or CMN syndrome) is associated with mosaic heterozygous activation mutations in *NRAS* with occasional extracutaneous involvement in large/giant CMN. However, *NRAS* mutation alone does not elicit a malignant transformation and loss of heterozygosity phenomena may be necessary for melanoma onset (1). Some studies have also shown *BRAF* mutations, especially in small CMN (13).

Despite a lack of data, it is accepted that only complete surgical excision is likely to negate the risk of melanoma in patients with CMN. This treatment is technically always feasible in small CMN and sometimes in medium-size CMN depending on location of the lesion. However, the benefit-to-harm ratio has to be carefully weighted as the risk of melanoma is probably very low in small-/medium-size CMN. In large/giant CMN, the melanoma risk is higher, but surgical excision, when feasible, is often only partial. Furthermore, melanoma can arise in subcutaneous sites or even in neurological or visceral sites in CMN syndrome. Thus, there is no evidence that surgery decreases the risk of melanoma for large/giant CMN, and the benefit-to-harm ratio is not always in favour of surgery in small-/medium-size CMN considering the likely very low risk of melanoma onset in these lesions.

In case of surgical abstention, clinical follow-up may be advised in order to diagnose melanomas early. In the current study, when surgical abstention was selected, the majority of physicians opted to follow up cases, and for this to occur at annual intervals from childhood. There is no data in the literature regarding whether follow-up is likely to decrease the risk of melanoma, and if it should start in childhood or adolescence/adulthood.

Concerning the aesthetic and psychological risk, the impact of CMN evolves with age, since morphological changes can appear over time with the development of papules, hair, verrucous appearance, ulcerations and benign proliferation nodules. Asymptomatic, CMN may usually be accompanied by pruritus, xerosis, and anhidrosis. The final colour of the naevus depends on the patient's phototype and is not related to the colour observed during the first months of life, with documented cases of spontaneous lightening of the naevus (14), especially on the scalp area (15). In a series of 29 large CMN, 25% of social problems and 30% of behavioural disorders are found in children. These disorders do not seem to correlate with the visibility of the lesion, its surgical treatment, or the age of the child, reflecting a general discomfort (16). Most parents (and children) with a CMN > 20 cm prefer a scar secondary to the treatment than the CMN itself, satisfaction being more important for the management of small CMN of the head and neck (17). Moreover, in a study by Bellier-Waast in 2008 (18), 55% of parents reported a feeling of rejection, 26% did not have pictures of their child before surgery, and the

psychological impact of the CMN was described as higher than that of the scar.

The profile of the sample of dermatologists in this study appears to be representative of the French Society of Dermatology membership. Of their members, 64% are female, 22% work in a hospital setting, and 78% work in private practice; figures that are comparable to the participants in the current study.

Although the sample of dermatologists in this study appears to be representative, there are significant differences between the number of representatives of each speciality in our study. Indeed, despite reminders, fewer paediatricians and surgeons responded to the survey, which may constitute a selection bias and impact on the results.

The clinical vignettes were chosen to represent each combination of potential clinical determinants, although the list of potential determinants was chosen *a priori* by 2 clinical experts, based on a non-systematic literature review and personal experience.

In conclusion, the clinician's level of experience in oncodermatology, their previous management of a melanoma on CMN, seniority, and a mixed case-load (paediatric and adult vs paediatric only) are determinants of whether a decision is taken to excise a CMN. This survey also confirms the variability of patient care at the national level in France. Thus, national recommendations to standardize clinical practices and the information provided to patients should be developed, involving several specialties to help guide a shared decision-making process. Moreover, sharing of experience between disciplines is useful, and thus joint consultations are interesting for decision-making.

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The authors have no conflicts of interest to declare.

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